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# BMJ Open

## Identifying Respiratory Health Research Priorities

Journal:	<i>BMJ Open</i>
Manuscript ID	bmjopen-2021-059326
Article Type:	Original research
Date Submitted by the Author:	15-Nov-2021
Complete List of Authors:	<p>Sharpe, Heather; University of Alberta, Medicine            Cerato, Lisa; Alberta Health Services            Derech, Darlene; Alberta Health Services            Guirguis, L; University of Alberta, Faculty of Pharmacy; University of Alberta,            Hayward, Kathleen; Alberta Health Services            Lohmann, Tara; University of Calgary, Medicine            MacLean, Joanna; University of Alberta, Pediatrics            Manafo, Elizabeth; Independent Consultant            Paskey, Janice; Alberta Health Services            Rasiah, Jananee ; University of Alberta            Rimkus, Mark; Alberta Health Services            Rizvi, Syeda ; Alberta Health Services            Robinson, Gerry; Alberta Health Services            Seefried, Brent; Alberta Health Services            Somani, Zeeyaan; Alberta Health Services            Tindall, Mindy; Alberta Health Services            Vliagoftis, Harissios; University of Alberta            Pendharkar, Sachin; University of Calgary, Medicine            Stickland , Michael K ; University of Alberta,</p>
Keywords:	RESPIRATORY MEDICINE (see Thoracic Medicine), Asthma < THORACIC MEDICINE, SLEEP MEDICINE

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Identifying Respiratory Health Research Priorities

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Abstract Word Count: 267

Manuscript Word Count: 2,787

Number of Figures: 1

Number of Tables: 3

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Identifying Respiratory Health Research Priorities

ABSTRACT

**Objective-** The Respiratory Health Strategic Clinical Network (RHSCN) was launched to facilitate respiratory and sleep health through implementation of innovative, patient-centred, evidence-informed coordinated services in Alberta. In collaboration with project partners, the RHSCN aimed to determine- what are the respiratory research priorities for Alberta? **Design-** The four phases research prioritization project included (1) identifying research questions from stakeholders, (2) determining which research questions had been answered in existing literature, (3) prioritizing unanswered questions, and (4) finalizing the priorities through an in-person workshop. **Setting-** The study occurred in Alberta, Canada, over a two-year period beginning in March 2017. **Participants-** A total of 446 individuals consented to participate in the survey. **Intervention-** the study included two stakeholder surveys. **Primary and Secondary Outcome Measures-** The Steering Committee designed the initial survey asking two open-ended core questions, (1) What questions do you have about breathing problems? And (2) What questions do you have about sleep problems? **Results-** A total of 595 possible questions were submitted, with 343 unique questions identified. Of the questions, 95 were out of scope, 155 answered by existing literature, and ten combined with others, while 83 were determined to be unanswered in the literature. Stakeholders were surveyed again to prioritize the remaining 83 questions, and they were reviewed by the project Steering Committee (clinicians and patients). At the in-person workshop, the Steering Committee identified 17 research topics as priority areas for respiratory and sleep research in Alberta. **Conclusion-** A stakeholder-led research prioritization process identified optimal clinical management/follow-up, equitable access to services, and management of social, psychological, mental health issues related to respiratory/sleep health as priority research areas.

## STRENGTHS AND LIMITATIONS OF THE STUDY

- Patients, caregivers, clinicians, and researchers identified the respiratory research priorities to assist in determining how research funding/support would be allocated, and to subsequently inform stakeholders of the chosen priorities.
- A four phase approach was used including (1) identifying research questions from stakeholders, (2) determining which research questions had been answered in existing literature, (3) prioritizing unanswered questions, and (4) finalizing the priorities through an in-person workshop.
- Two open-ended core questions were asked of survey participants (1) What questions do you have about breathing problems? And (2) What questions do you have about sleep problems?

Identifying Respiratory Health Research Priorities

INTRODUCTION

In Alberta, respiratory disease is a prevalent, costly burden on the health care system. It is estimated that chronic obstructive pulmonary disease (COPD) costs the province over \$250 million per year[1], while asthma leads to missed days at school, frequent health care utilization, and negative impacts on families' quality of life.[2]–[4] Obstructive sleep apnea (OSA) is likely underdiagnosed in Canada[5] and may lead to reduced quality of life, workplace and vehicular accidents, and decreased work productivity[6]–[10]. In early 2014 the Respiratory Health Strategic Clinical Network (RHSCN) was launched by Alberta Health Services (the provincial health authority) to address the many challenges associated with respiratory and sleep disorders with the aim to facilitate optimal respiratory health through implementation of innovative, patient-centred, evidence-informed and coordinated services in the province of Alberta.[11] The RHSCN supports all respiratory-related conditions, with priority areas being asthma, COPD, and sleep-disordered breathing. The RHSCN consists of a Core Committee, which guides the overall direction of the Network, as well as several working groups that undertake specific projects to support the provision of evidence-based respiratory health care within the Alberta Health Services through quality improvement initiatives, development of clinical practice guidelines and implementation of innovative interventions. The scientific office within the RHSCN facilitates clinical research through funding and research support to promote clinical respiratory research, uptake of best evidence, building research capacity in the province and directs the Network's research priorities.

Historically, research agendas were dictated by the scientific community; however, there is a growing recognition that patients, caregivers, and clinicians should be engaged in identifying research priorities.[12] Increasingly in Canada there have been several research prioritization activities that used a patient-oriented approach.[13]–[15] The James Lind Alliance (JLA) suggests that patients, caregivers and clinicians work together to identify treatment uncertainties related to specific conditions and

prioritize those they collectively agree to be most important.[16] Using a modified JLA approach, the purpose of this project was to engage patient, caregiver, clinician, and researcher stakeholders to identify the respiratory research priorities that would assist the RHSCN scientific office in determining how research funding/support would be allocated, and to subsequently inform stakeholders of the chosen priorities.

## **METHODS**

### **Steering Committee Development**

The project was conducted from March 2017 to February 2019. Project oversight was performed by a Steering Committee. Patient engagement professionals from Alberta Health Services and Alberta Strategy for Patient-Oriented Research Support for People and Patient-Oriented Research and Trials Unit (AbSPORU) helped to identify patients/caregivers who might be interested in Steering Committee membership. Patients and caregivers met with the project lead (HS) by phone to discuss the scope of the project and required commitment. Steering Committee members were not compensated beyond travel expenses. The steering committee was comprised of four individuals with lived experience, two caregivers, four respiratory physicians, two respiratory therapists, two pharmacists, one nurse and the Scientific Director of the RHSCN; within the group there was representation from academic and community-based clinicians. We did not request Steering Committee members to disclose personal information related to dimensions of diversity, however the group acknowledged the importance of broad representation, felt the group was generally representative, while respecting the privacy of the team members.

### **Priority Setting Process**

The initial in-person meeting of the Steering Committee provided an opportunity to introduce the project, discuss examples of similar work from other groups,[14] and review the process of priority



setting. As the RHSCN addresses both respiratory and sleep health concerns, both topics were included in the priority setting and deemed equally important.

The Steering Committee chose to follow a modified James Lind Alliance (JLA) Priority Setting Partnership method, as described by The Alberta Depression Research Priority Setting Project.[14] This approach was selected as it had demonstrated feasibility, rigor, and significant patient/caregiver engagement[17]. The adopted process was similar to the JLA method, however the voices of individuals with lived experience participated throughout (without the funnel approach, whereby the number of participants are reduced at each phase) with an egalitarian, consensus-building strategy within the Steering Committee [13]. The four-phase process included (1) identifying potential research questions by stakeholders, (2) determining research questions that had been answered, (3) prioritizing unanswered questions, and (4) finalizing the priority list at an in-person workshop.

**Participant Recruitment**

Study participants included individuals that participated in the online surveys designed to elicit their research priorities. Participants of the survey targeted three groups: (1) individuals with lived experience of respiratory disease or sleep disorders, (2) caregivers of individuals with respiratory disease or sleep disorders, and (3) health care providers to individuals with respiratory disease or sleep disorders. Participants self-identified to which group(s) they belonged via the survey. Posters and cards with QR codes and website links were disseminated through stakeholders, such as respiratory and sleep clinics and project partners, and all were asked to share widely. Steering Committee members were also asked to share the flyer within their community (such as support groups, online forums, professional groups etc.) and through social media platforms. The RHSCN further shared the survey links through provincial respiratory/sleep newsletters and email communications to members. Participants were not directly reimbursed but offered to enter a draw for a gift card prize for participation.

## Patient and Public Involvement

This project was co-led by patient/caregiver team members of the Steering Committee. Every phase of the methodology included patient/caregiver involvement, as they were equal members and active decision makers. Patients and caregivers were involved in the study design, implementation, data analysis and manuscript preparation (including authorship). Additionally, knowledge translation activities will include working with patient/family groups to disseminate the research prioritization questions.

## Ethics Approval

Ethics approval was received by the health research ethics board of the University of Alberta (Pro00075354).

## DATA ANALYSIS

### Phase 1: Identify Potential Research Questions by Stakeholders

In order to obtain broad input of potential research questions, the Steering Committee designed an initial survey asking two open-ended questions:

1. What questions do you have about breathing problems?
2. What questions do you have about sleep problems?

Participants were provided with rationale and context for the survey, information about how the data would be used, contact information for seeking further information and asked to provide their consent (by clicking a consent box on the online survey) to participate and use their responses for the study.

They were invited to answer one or both of the questions. They were also asked optional questions including gender and age group. The survey was designed to take less than ten minutes to complete and was first reviewed/tested by members of the Steering Committee with lived experience. The survey was completed online over a two-month period using the Research Electronic Data Capture (REDCap)

online platform, hosted by the University of Calgary. REDCap is a secure, web-based software platform designed to support data capture for research studies.[18], [19] Incomplete surveys (those without submitted questions) were excluded from the analysis.

**Phase 2: Determine Research Questions that Had Been Answered**

Appendix A provides a detailed summary of the process used to identify whether the submitted questions had already been answered in the literature. In brief, research questions that were submitted by participants via the survey in Phase 1 were reviewed by two members of the team. Duplicate questions were removed and similar questions were collated into broad topics. Questions were reframed where necessary to ensure they identified the patient population, intervention, control and outcome (PICO) where possible. For each question topic, the following databases were searched for relevant, up-to-date systematic reviews, clinical practice guidelines and/or study protocols (i.e., studies in process):

- Cochrane Database of Systematic Reviews (EBM Reviews)
- American College of Physicians (ACP) Journal Club via Medline (Ovid)
- Database of Abstracts of Reviews of Effects (DARE) via Medline (Ovid)
- Medline databases (Ovid)
- Canadian Respiratory Guidelines (Canadian Thoracic Society)
- Global Initiative for Asthma (GINA)
- AAFP (USA)
- NICE guidelines (UK)

Questions that were determined not to be adequately answered in the literature (i.e. the topic was not addressed by the above sources) were included in the list of unanswered questions to move forward to

Phase 3, while questions confirmed to have been already answered did not move forward to Phase 3. Although policy and knowledge translation questions were initially considered out of scope, they were provided to the Steering Committee in a separate list for their consideration in Phase 3. This was done at the request of the Steering Committee as the group felt that knowledge translation and policy questions may have significant relevance to the work of the RHSCN.

### **Phase 3: Prioritization of Unanswered Questions**

The final list of unanswered questions was divided into two categories: 1) questions relating to breathing problems and 2) questions relating to sleep problems. Participants in Phase 1 were contacted again and asked to select their top ten priorities from one or both categories via a new RedCAP survey. Data from this survey was used to inform the Steering Committee's discussion during the final workshop.

### **Phase Four: Final In-Person Priority Setting Workshop**

The Steering Committee participated in an in-person workshop with additional participation by health professional groups and patient/caregivers that were underrepresented, such as pharmacy and parents/guardians, with a total of 11 participants. The workshop was led by a trained facilitator with experience working with the Strategic Clinical Networks and an understanding of Alberta's health care system. The workshop included an overview of the results of the two surveys, and the RHSCN scope and work. Following a discussion of the results, the Steering Committee participants split into two groups to prioritize the existing sleep research questions and respiratory research questions. The two groups compared priority rankings, removing questions that were deemed as low priority by both groups. The Steering Committee discussed the remaining questions and subsequently conducted a second round of prioritization, again removing low priority questions. The final list of questions was determined through group consensus, and included combining some questions when appropriate and refining wording.

During the workshop, participants made the following key decisions:

1. The list should not be guided by an arbitrary number (e.g., top ten); rather, the most important questions should be included in the list.
2. Several ‘out of scope’ questions were important and should not be dismissed without consideration, despite this approach being atypical from research prioritization methods
3. The significant overlap between respiratory and sleep health created redundancies in two lists, and a hybrid approach was developed.

**RESULTS**

A total of 461 unique individuals accessed the first survey (Table 1). Fifteen participants’ responses were removed as they did not clearly provide consent, leaving a total of 446 survey responses. The survey methods were successful at gaining broad representation from the three core groups: 317 participants stated they were an individual with lived experience with respiratory disease/patient (71.1%), 207 stated they were a caregiver of an individual with respiratory disease (46.4%) and 276 stated they were a health care professional that provided care for individuals with respiratory disease (61.9%). Of note, many respondents selected more than one category with which they identified.

A total of 595 research questions were submitted (see Figure 1). Two data scientists reviewed the questions for redundancy, leaving 343 unique questions. Of the remaining questions, 66 were determined to be answered in the existing literature while 94 were judged to be out of scope (policy and cost related questions). Of the 183 research questions remaining, five questions were deemed not to be research questions, 86 were mapped to evidence and identified as answered, while 92 were identified as unanswered, and remained in the list of unanswered questions for consideration. The Steering Committee determined that of the 92 questions, three questions were answered in the literature, one question was determined to be out of scope, and five were combined with other questions or deemed

not to be research questions, leaving 83 possible research questions. Table 2 provides an overview of the mapping of the final research questions and answered questions by category.

One hundred forty stakeholders participated in the second survey to rank the remaining questions. The in-person workshop allowed the Steering Committee to review the remaining questions collectively and identify 17 final priority areas for respiratory and sleep research in Alberta. The final list included six questions for sleep health, three questions for respiratory health, and eight questions that were applicable to both sleep and respiratory health (see Table 3).

## DISCUSSION

The RHSCN Research Prioritization process identified 17 priorities for respiratory and sleep health, integrating the perspectives of patients/caregivers and clinicians/researchers. This research is novel as it was co-owned and co-developed by patient/caregivers and clinicians/researchers, and is the first Canadian research prioritization project that aimed to address both respiratory and sleep health. This prioritization will assist with decision making for the RHSCN related to allocation of resources and Network priorities.

Including individuals with lived experience as co-owners in research increases legitimacy and opportunity for innovation.[20] Additionally, researchers have moral and ethical responsibilities to ensure that the voice of patients and families are represented in research priorities.[21] A strength of this research is the high participation of patients and caregivers in the process, which is reflected in the nature of the research questions prioritized. As suggested by Breault and colleagues[14], the higher the level of participation from individuals with lived experiences (and their caregivers), the more closely a research prioritization list will reflect the true priorities of the community. The questions identified in this study had a strong focus on policy, equitable access to services, and the management of social, psychological, mental health issues related to respiratory/sleep health. Over two-thirds of survey

respondents indicated they were patients, and almost half stated they were caregivers of someone with a respiratory or sleep problem (individuals could select more than one role).

Respiratory specific research prioritization has also increased in prominence. In the Netherlands, patient focus groups and questionnaires were used to identify research priorities for asthma and COPD.[22] Two significant priorities that arose were knowledge about the causes of the diseases and development of more effective medications, with little emphasis upon socio-political factors that impact health care. In contrast, our priorities for respiratory/sleep health research had a strong emphasis on social, economic and health care access issues. This variance may be the result of the autonomy of the Steering Committee, who made modifications to the proposed structure and inclusion criteria of the priorities by including out of scope policy and economic questions in the final review. Lastly, this discrepancy may also be a reflection of shifting priorities over time, and the nature of the RHSCN being focused on health service delivery, rather than basic science research. A JLA initiative is currently underway in Saskatchewan, Canada to assess the research priorities for sleep apnea.(19) In the Canadian province of Quebec, a survey of 148 patients with a more narrow scope of COPD only determined that relief of breathlessness was a research and health care priority for patients.(20)

**LIMITATIONS**

There are limitations to consider related to this research. First, an online survey platform was convenient, cost-effective and timely, however it may have prevented some individuals from participating in the research. There was general concern that individuals living with COPD may be underrepresented, based upon their age, access to online resources and ability to complete the survey. However, given several important COPD specific questions that were raised in the survey, this voice may have been well represented. Second, we did not undertake specific measures to target Indigenous populations and caregivers, those with a primary language other than English, as well as those with limited internet access. In future research, we would advocate for requesting ethnicity/race information

from participants and using this information to guide recruitment of participants. As well, we would suggest developing a targeted strategy for ensuring representation from those without internet access, and ensuring key populations are included. Third, the time commitment for participating as a Steering Committee member was substantial. Members attended two full-day in-person meetings and approximately six hours of telephone meetings, plus numerous emails/material review over the course of 18 months. The participation level of the patient/caregiver steering committee demonstrated strong commitment, however future groups may consider additional strategies, such as role-sharing, to decrease volunteer fatigue. Finally, this research focused on the province of Alberta, and while the results may be broadly translatable, generalizability may be limited. Future results from other prioritization activities in other jurisdictions may help to validate our findings.

## CONCLUSION

By involving individuals with lived experience with respiratory disease, caregivers and health care professionals, a research priority list comprising six questions for sleep health, three questions for respiratory health, and eight questions that were applicable to both sleep and respiratory health was developed. This research exercise will lead to a more focused distribution of research funds and other resources locally, and will inform other groups as they look to support research that is more relevant to patients.



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**Acknowledgements:** Our sincere appreciation to the Respiratory Health Section of the Medicine Strategic Clinical Network Core Committee for their ongoing support of this project. In addition, we thank all of the participants in the surveys and the research prioritization process.

**Funding statement:** This research received no specific grant from any funding agency in the public, commercial or not-for-profit sectors. This work was supported by an in-kind contribution by the Respiratory Health Strategic Clinical Network™ of Alberta Health Services and the Alberta SPOR SUPPORT Unit Patient Engagement Platform.

**Conflict of Interest Statement:** Ms. Hayward received honoraria for speaking at continuing education activities for health care professionals from Astra Zeneca in the last year, unrelated to this project. Dr. Sachin Pendharkar discloses he has received advisory fees from Jazz Pharmaceuticals, Paladin Labs; received research funding from Canadian Institute for Health Research, Alberta Health Services, The Lung Association, and MITACS; additionally, he has participated in a study by Jazz Pharmaceuticals (site co-investigator with no personal stipend or salary). Dr. Heather Sharpe & Dr. Michael Stickland received research funding from the Alberta Boehringer Ingelheim Collaboration and Dr. Sharpe is a member of the Board of Directors of the Canadian Network of Respiratory Care. The remaining authors have no other disclosures.

**Availability of data and materials-** the datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

**Ethics approval-** granted from the Health Research Ethics Board at the University of Alberta.

**Contributors’ Statement**

\*As this work was co-owned and developed in collaboration with our patient/caregiver advisors, all members of the Steering Committee were responsible for study design and interpretation of results. All authors have given permission for the submission of the manuscript, and have agreed to be listed as a co-author.

Heather Sharpe –led the Steering Committee (SC) through the study design process, data collection and analysis. Was responsible for drafting the manuscript, editing and is corresponding author.

Lisa Cerato-facilitated the final workshop to the creation of the final prioritization list. Provided feedback on the manuscript.

Darlene Derech-a patient/caregiver advisor that was a member of the SC. Involved in the study design, data collection, analysis and final research prioritization. Was responsible for providing feedback on the manuscript.

Lisa Guirguis-a health professional/academic representative on the SC. Involved in the study design, data collection, analysis and final research prioritization. Was responsible for providing feedback on the manuscript.

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11 Elizabeth Manafo-conducted the determination if submitted research questions were answered in the  
12 existing literature and responsible for the methodology and reporting. Provided feedback on the  
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15 Janice Paskey - a patient/caregiver advisor that was a member of the SC. Involved in the study design,  
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Michael K Stickland- the Scientific Director of the RHSCN. Involved in the study design, data collection, analysis and final research prioritization. Was responsible for providing feedback on the manuscript.

For peer review only

Table 1. Survey participant demographic characteristics

Characteristic	No (%) of total respondents (n=448)
<b>Do you prefer to answer questions related to breathing problems, sleep problems or both? (n=409)</b>	
Breathing problems	81 (19.8)
Sleep problems	123 (30.1)
Both (breathing and sleep problems)	205 (50.1)
<b>Category-may choose more than 1 (n=446)</b>	
Person with breathing problems	132 (29.6)
Person with sleep problems	185 (41.5)
Family/caregiver of person with breathing problems	90 (20.2)
Family/caregiver of person with sleep problems	117 (26.2)
Health care professional-breathing problems	148 (33.2)
Health care professional-sleep problems	128 (28.7)
Other (dentist, scientist, patient with co-morbidities)	11 (2.5)
<b>Gender (n=446)</b>	
Female	347 (77.8)
Male	86 (19.3)
Non-Binary	5 (1.1)
Prefer not to respond/Did not respond	8 (1.8)
<b>Age</b>	
≤17 years	1 (.2)
18-29 years	44 (9.9)
30-49 years	116 (26.0)
50-79 years	197 (44.2)
≥80 years	4 (.9)
Did not respond	4 (.9)

Table 2. Submitted Research Questions by Topic Reviewed and Finalized by Steering Committee

Total	Total questions	Answered in Literature	Remaining Research questions
Asthma	33	20	13
Combustibles	4	1	3
COPD	3	2	1
Devices	13	4	9
Diagnostic	7	4	3
General Medicine	14	11	3
ILD	9	4	5
Nasal	4	3	1
OSA	25	19	6
Sleep	35	17	18
Other	25	4	21
Cost and policy	0	0	0
Total	172	89	83

\*cost and policy questions were deemed out of scope and represented in the out of scope data

Table 3. List of Final Research Questions

<b>Questions for Respiratory and Sleep</b>	
1.	What is the relationship between social, psychological, and mental health issues and respiratory disease/sleep disorders for the individual/the family?
2.	What is the economic impact of respiratory disease/sleep disorders for community and inpatient/urgent care in Alberta?
3.	For individuals with suspected respiratory disease/sleep disorders, when is it appropriate for a primary care provider to refer to a specialist, compared to continuing care, to ensure the best treatment/health outcomes? What are the educational needs of primary care providers to facilitate this?
4.	What model of care is most effective at transitioning patients and their families from paediatric to adult respiratory/sleep care?
5.	For individuals living with respiratory disease/sleep disorders, what are the most effective self-management interventions and/or community supports/resources?
6.	What strategies will improve equitable access to respiratory/sleep care for Albertans?
7.	What are the patient & families' priorities related to the treatment of their breathing/sleep problems?
8.	For individuals with respiratory/sleep problems, which interventions, resources, programs in the community will result in fewer specialty care, emergency department or hospital visits?
<b>Questions for Respiratory</b>	
1.	How can we improve access to pulmonary rehabilitation for individuals with respiratory disease?
2.	For individuals with respiratory disease, how do environmental factors (humidity, air pollution etc.) impact disease control/management in Alberta?
3.	How does an individual with respiratory disease determine if different therapies will be worth the additional cost (money, risk of side effects, exacerbations)?
<b>Questions for Sleep</b>	
1.	For individuals with suspected sleep-related breathing disorders (e.g. obstructive sleep apnea), what is the recommended ongoing clinical management/follow-up care to improve and sustain health outcomes?
2.	Do treatments besides CPAP improve outcomes for individuals with sleep-related breathing disorders (e.g. obstructive sleep apnea and/or hypoventilation)?
3.	What is the current quality of provision and interpretation of investigations for sleep-related breathing disorders in Alberta?
4.	What strategies can be used to promote sleep as an important contributor to health?
5.	For individuals experiencing poor quality sleep, at what point should they seek medical advice to improve their health outcomes?
6.	What are the policy factors that inform insurable coverage for testing and treatment of sleep-related breathing disorders in Alberta?

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Appendix A. Verification Process to Identify Uncertainties

Where, no up-to-date, reliable systematic reviews or guidelines of research evidence addressing the uncertainty exist/have not been adequately addressed. If a study protocol exists, this will be noted. These uncertainties will move forward for interim prioritization.

Process	Description	Output
a. Check evidence-base for relevant, up to date systematic reviews, guidelines and study protocols (i.e., studies in process)	<ul style="list-style-type: none"><li>i. Cochrane Database of Systematic Reviews (EBM Reviews)</li><li>ii. American College of Physicians (ACP) Journal Club via Medline (Ovid)</li><li>iii. Database of Abstracts of Reviews of Effects (DARE) via Medline (Ovid)</li><li>iv. Medline databases (Ovid)</li><li>v. <a href="#">Canadian Respiratory Guidelines</a> (Canadian Thoracic Society)</li><li>vi. <a href="#">Global Initiative for Asthma</a> (GINA)</li><li>vii. <a href="#">AAFP</a> (USA)</li><li>viii. <a href="#">NICE guidelines</a> (UK)</li><li>ix. Other (used if unique population, intervention or outcome would be better addressed in a different database)</li></ul>	If evidence is available, cite it with primary author conclusion.
	Using specific search terms as they relate to the submission topic area, subtopic area and outcome (as identified). Specific search outputs will be documented as they relate to each topic and subtopic area.	
	To check questions about sleep and breathing risk factors, which are mainly addressed by reviews including prospective cohort studies, other systematic reviews. MEDLINE, EMBASE, CINAHL and PsycINFO will be searched with the same keywords using a systematic review filter.	
b. Determine if evidence is reliable and relevant using	1] Systematic review	
	1.1 Has it been published in the last 3 years?	[YES > does not move forward; NO > moves forward]

Process	Description	Output
the following criteria:	<b>AND</b> 1.2 For evidence not derived from Cochrane et al., ask the following: Does it have published clear methods? <ul style="list-style-type: none"> <li>• Where inclusion/exclusion criteria reported?</li> <li>• Was search adequate?</li> <li>• Where the included studies synthesized?</li> <li>• Was the quality of included studies assessed?</li> <li>• Are sufficient details about the individual included studies presented?</li> <li>• Was any potential bias addressed/managed?</li> </ul>	[YES > does not move forward; NO > moves forward]
	<b>AND</b> 1.3 Are reported findings clinically significant?	[YES > does not move forward; NO > moves forward]
	<b>2] Guidelines</b>	
	2.1 Are guidelines evidence-based, that are used as a framework for clinical decision making and supporting best practices from authoritative Canadian, American or UK bodies?	[YES > does not move forward; NO > moves forward]
	<b>3] Protocol</b>	
	3.1 If no systematic review or guidelines have been located, conduct Protocol search through EBM Cochrane	[YES > note its existence; NO > moves forward]
		If <b>all of the criteria</b> have been answered with a <b>Yes = "Known unknown"</b> [tier 2]  If <b>one or more</b> of the criteria has been answered with a <b>No = Uncertainty</b>

Process	Description	Output
c. Verify selection output	Clinical significance must be determined by Steering Committee based on reported findings and recorded author conclusions.	Long-list of uncertainties are identified for interim prioritization (and 'known unknowns' are recorded with evidence cited)

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### Reporting guideline for health research priority setting with stakeholders (REPRISE)

No	Item	Descriptor and/or examples
A	Context and scope	
1	Define geographical scope	Provincial scope
2	Define health area, field, focus	Respiratory and sleep health
3	Define the intended beneficiaries	Individuals living with respiratory and sleep disorders in the province of Alberta and their families.
4	Define the target audience of the priorities	Respiratory Health Section of the Medicine Strategic Clinical Network-to use the prioritization to fund research
5	Identify the research area	Health service research and clinical research
6	Identify the type of research questions	Was not pre-defined
7	Define the time frame	To provide research prioritization for five years
B	Governance and team	
8	Describe the selection and structure of the leadership and management team	The Scientific Office of the Network was primarily responsible for the development of the Steering Committee. The Steering Committee was created to ensure a strong voice for those with lived experience of respiratory and/or sleep disorders. The Core Committee of the Network provided content expertise.
9	Describe the characteristics of the team	Project oversight was performed by a Steering Committee comprising four individuals with lived experience, two caregivers, four respiratory physicians,

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No	Item	Descriptor and/or examples
		two respiratory therapists, two pharmacists, one nurse and the Scientific Director of the RHSCN, within the group there was representation from academic and community based clinicians. We did not request Steering Committee members to disclose sex/gender or race/ethnicity, however the group was diverse in perceived age, sex, gender, ethnicity and academic/non-academic background.
10	Describe any training or experience relevant to conducting priority setting	The Patient Engagement Platform of the Alberta Strategy for Patient-Oriented Research's Support for People and Patient-Oriented Research and Trials Unit provided guidance, a consultant with priority setting experience was hired to assist with identifying unanswered questions and a facilitator was brought to the team for the final priority setting workshop.
C	Framework for priority setting	
11	State the framework used (if any)	A modified James Lind Alliance approach was used based upon the Alberta Depression Research Priority Setting Project
D	Stakeholders or participants	
12	Define the inclusion criteria for stakeholders involved in priority-setting	Patients, caregivers, health professionals, researchers, policy makers, and other stakeholders that self-identified as having an interest in respiratory/sleep health
13	State the strategy or method for identifying and engaging stakeholders	Posters and cards with QR codes and website links were disseminated through stakeholders, such as respiratory and sleep clinics. Steering Committee members were asked to share the flyer within their community (such as support groups, online forums etc.) and through social media platforms. The

No	Item	Descriptor and/or examples
		RHSCN shared the survey links through newsletters and email communications to members.
14	Indicate the number of participants and/or organizations involved	A total of 461 unique individuals accessed the first survey. Fifteen participants' responses were removed as they did not provide clear consent, leaving a total of 446 survey responses.
15	Describe the characteristics of stakeholders	Three core groups: 317 participants stated they were an individual with lived experience with respiratory disease/patient (71.1%), 207 stated they were a caregiver of an individual with respiratory disease (46.4%) and 276 stated they were a health care professional that provided care for individuals with respiratory disease (61.9%). Of note, many respondents selected more than one category with which they identified.
16	State if reimbursement for participation was provided	Individuals that participated on the Steering Committee were provided reimbursement for any expenses associated with attending the in-person meetings including travel (hotel, mileage, parking, meals etc.). Participants were not paid an honoraria or other compensation. Participants in the survey were invited to enroll in a draw for a small gift card to a book store for participating.
E	Identification and collection of research priorities	
17	Describe methods for collecting initial priorities	The four phases to the project included (1) identifying research questions by stakeholders, (2) determining research questions that had been answered in existing literature (guidelines, systematic reviews etc.), (3) prioritization of unanswered questions, and (4) finalization through an in-person priority setting workshop. The Steering Committee designed the initial survey asking



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No	Item	Descriptor and/or examples
		two open-ended core questions, (1) What questions do you have about breathing problems? And (2) What questions do you have about sleep problems?
18	Describe methods for collating and categorizing priorities	Priorities were originally organized into the broad categories of respiratory health and sleep health. Subsequently they were categorized into sub-groups to facilitate data management. For example, respiratory health priorities were organized into: asthma, COPD, medications, devices, obstructive sleep apnea, cost & policy, diagnostic, nasal, sleep, interstitial lung disease, and other.
19	Describe methods and reasons for modifying (removing, adding, reframing) priorities	All proposed priorities were reframed as necessary to create a PICO (patient, intervention, control & outcome) question were possible. Priorities that overlapped were combined.
20	Describe methods for refining or translating priorities into research topics or questions	Two members of the team were responsible for creating research questions from the submissions. The resulting list was reviewed by the Steering Committee.
21	Describe methods for checking whether research questions or topics have been answered	For each question topic, key databases were searched for relevant, up-to-date systematic reviews, clinical practice guidelines and/or study protocols (i.e., studies in process). Questions that had not been adequately answered in the literature (the topic was not addressed by the above sources), were to be included in the list of unanswered questions. Questions that were answered in the literature were identified as 'known unknowns', and along with possibly out of scope questions (policy questions or those that would not be answered by the literature sources) were included by the two reviewers in a separate list for review by list to the Steering Committee. Questions for final inclusion

No	Item	Descriptor and/or examples
		based on clinical and research significance were determined by the Steering committee.
22	Describe number of research questions or topics	A total of 595 research questions were submitted (see Figure 1). A team of two people identified answers for 343 (74%) questions within the literature. A total of 178 research questions were identified as unanswered questions that moved forward. One hundred forty stakeholders participated in the second survey to rank the remaining questions. A final list of 17 research topics was identified as priority areas for respiratory and sleep research in Alberta through this prioritization.
F	Prioritization of research topics/questions	
23	Describe methods and criteria for prioritizing research topics or questions	Two online surveys were completed. The first, to identify possible priorities, the second to prioritize the identified priorities from the first survey. The Network Core Committee reviewed the results and provided their input to the Steering Committee. An in person priority setting workshop was completed to identify the final priorities.
24	State the method or threshold for excluding research topics/questions	A consensus approach was used. The Steering Committee reached consensus on what questions would be included in the final list.
G	Output	
25	State the approach to formulating the research priorities	PICO (population, intervention, comparator, outcome) questions were formulated, however the Steering Committee did revise the priorities in the final priority setting workshop.
H	Evaluation and feedback	

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No	Item	Descriptor and/or examples
26	Describe how the process of prioritization was evaluated	The prioritization process has not yet been formally evaluated, however the impact will be assessed through the Scientific Office of the Network.
27	Describe how priorities were fed back to stakeholders and/or to the public; and how feedback (if received) was addressed and integrated	The results of the priority setting were shared with the RHSCN, including the Core Committee and the working groups.
I	Implementation	
28	Outline the strategy or action plans for implementing priorities	The priorities have been used to guide the funding of research grants offered by the Network, including seed grant funding and studentships.
29	Describe plans, strategies, or suggestions to evaluate impact	The Network evaluates the impact of the scientific office annually, as well as is involved in the evaluation and priority setting for the Network every 3-5 years.
J	Funding and conflict of interest	
30	State sources of funding	This work was supported by an in-kind contribution by the Respiratory Health Strategic Clinical Network™ of Alberta Health Services and the Alberta SPOR SUPPORT Unit Patient Engagement Platform.
31	Declare any conflicts or competing interests	The authors have no other disclosures.

### Checklist for Reporting Results of Internet E-Surveys (CHERRIES)

<b>Checklist Item</b>	<b>Explanation</b>	<b>Page Number</b>
Describe survey design	Describe target population, sample frame. Is the sample a convenience sample? (In “open” surveys this is most likely.)	Page 6-7
IRB approval	Mention whether the study has been approved by an IRB.	Page 7
Informed consent	Describe the informed consent process. Where were the participants told the length of time of the survey, which data were stored and where and for how long, who the investigator was, and the purpose of the study?	Page 7
Data protection	If any personal information was collected or stored, describe what mechanisms were used to protect unauthorized access.	n/a
Development and testing	State how the survey was developed, including whether the usability and technical functionality of the electronic questionnaire had been tested before fielding the questionnaire.	Page 7
Open survey versus closed survey	An “open survey” is a survey open for each visitor of a site, while a closed survey is only open to a sample which the investigator knows (password-protected survey).	Page 7
Contact mode	Indicate whether or not the initial contact with the potential participants was made on the Internet. (Investigators may also send out questionnaires by mail and allow for Web-based data entry.)	Page 7
Advertising the survey	How/where was the survey announced or advertised? Some examples are offline media (newspapers), or online (mailing lists – If yes, which ones?) or banner ads (Where were these banner ads posted and what did they look like?). It is important to know the wording of the announcement as it will heavily influence who chooses to participate. Ideally the survey announcement should be published as an appendix.	Page 6
Web/E-mail	State the type of e-survey (eg, one posted on a Web site, or one sent out through e-mail). If it is an e-mail survey, were the responses entered manually into a database, or was there an automatic method for capturing responses?	Page 6
Context	Describe the Web site (for mailing list/newsgroup) in which the survey was posted. What is the Web site about, who is visiting it, what are visitors normally looking for? Discuss to what degree the content of the Web site could pre-select the sample or influence the results. For example, a survey about vaccination on a anti-immunization Web site will have different results from a Web survey conducted on a government Web site	Page 6
Mandatory/voluntary	Was it a mandatory survey to be filled in by every visitor who wanted to enter the Web site, or was it a voluntary survey?	Page 6
Incentives	Were any incentives offered (eg, monetary, prizes, or non-monetary incentives such as an offer to provide the survey results)?	Page 6

Time/Date	In what timeframe were the data collected?	Page 6
Randomization of items or questionnaires	To prevent biases items can be randomized or alternated.	n/a
Adaptive questioning	Use adaptive questioning (certain items, or only conditionally displayed based on responses to other items) to reduce number and complexity of the questions.	n/a
Number of Items	What was the number of questionnaire items per page? The number of items is an important factor for the completion rate.	Page 7
Number of screens (pages)	Over how many pages was the questionnaire distributed? The number of items is an important factor for the completion rate.	Page 7
Completeness check	It is technically possible to do consistency or completeness checks before the questionnaire is submitted. Was this done, and if “yes”, how (usually JavaScript)? An alternative is to check for completeness after the questionnaire has been submitted (and highlight mandatory items). If this has been done, it should be reported. All items should provide a non-response option such as “not applicable” or “rather not say”, and selection of one response option should be enforced.	n/a
Review step	State whether respondents were able to review and change their answers (eg, through a Back button or a Review step which displays a summary of the responses and asks the respondents if they are correct).	n/a
Unique site visitor	If you provide view rates or participation rates, you need to define how you determined a unique visitor. There are different techniques available, based on IP addresses or cookies or both.	n/a
View rate (Ratio of unique survey visitors/unique site visitors)	Requires counting unique visitors to the first page of the survey, divided by the number of unique site visitors (not page views!). It is not unusual to have view rates of less than 0.1 % if the survey is voluntary.	n/a
Participation rate (Ratio of unique visitors who agreed to participate/unique first survey page visitors)	Count the unique number of people who filled in the first survey page (or agreed to participate, for example by checking a checkbox), divided by visitors who visit the first page of the survey (or the informed consents page, if present). This can also be called “recruitment” rate.	n/a
Completion rate (Ratio of users who finished the survey/users who	The number of people submitting the last questionnaire page, divided by the number of people who agreed to participate (or submitted the first survey page). This is only relevant if there is a separate “informed consent” page or if the survey goes over several pages. This is a measure for attrition. Note that	n/a

agreed to participate)	"completion" can involve leaving questionnaire items blank. This is not a measure for how completely questionnaires were filled in. (If you need a measure for this, use the word "completeness rate".)	
Cookies used	Indicate whether cookies were used to assign a unique user identifier to each client computer. If so, mention the page on which the cookie was set and read, and how long the cookie was valid. Were duplicate entries avoided by preventing users access to the survey twice; or were duplicate database entries having the same user ID eliminated before analysis? In the latter case, which entries were kept for analysis (eg, the first entry or the most recent)?	n/a
IP check	Indicate whether the IP address of the client computer was used to identify potential duplicate entries from the same user. If so, mention the period of time for which no two entries from the same IP address were allowed (eg, 24 hours). Were duplicate entries avoided by preventing users with the same IP address access to the survey twice; or were duplicate database entries having the same IP address within a given period of time eliminated before analysis? If the latter, which entries were kept for analysis (eg, the first entry or the most recent)?	n/a
Log file analysis	Indicate whether other techniques to analyze the log file for identification of multiple entries were used. If so, please describe.	n/a
Registration	In "closed" (non-open) surveys, users need to login first and it is easier to prevent duplicate entries from the same user. Describe how this was done. For example, was the survey never displayed a second time once the user had filled it in, or was the username stored together with the survey results and later eliminated? If the latter, which entries were kept for analysis (eg, the first entry or the most recent)?	n/a
Handling of incomplete questionnaires	Were only completed questionnaires analyzed? Were questionnaires which terminated early (where, for example, users did not go through all questionnaire pages) also analyzed?	Page 8
Questionnaires submitted with an atypical timestamp	Some investigators may measure the time people needed to fill in a questionnaire and exclude questionnaires that were submitted too soon. Specify the timeframe that was used as a cut-off point, and describe how this point was determined.	n/a
Statistical correction	Indicate whether any methods such as weighting of items or propensity scores have been used to adjust for the non-representative sample; if so, please describe the methods.	n/a

This checklist has been modified from Eysenbach G. Improving the quality of Web surveys: the Checklist for Reporting Results of Internet E-Surveys (CHERRIES). J Med Internet Res. 2004 Sep 29;6(3):e34 [erratum in J Med Internet Res. 2012; 14(1): e8.]. Article available at <https://www.jmir.org/2004/3/e34/>; erratum available <https://www.jmir.org/2012/1/e8/>. Copyright ©Gunther Eysenbach. Originally published in the [Journal of Medical Internet Research](https://www.jmir.org/2004/3/e34/), 29.9.2004 and 04.01.2012.

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# BMJ Open

## What Are the Respiratory Health Research Priorities in Alberta Canada? A Stakeholder Consultation

Journal:	<i>BMJ Open</i>
Manuscript ID	bmjopen-2021-059326.R1
Article Type:	Original research
Date Submitted by the Author:	16-Apr-2022
Complete List of Authors:	Sharpe, Heather; University of Alberta, Medicine Cerato, Lisa; Alberta Health Services Derech, Darlene; Alberta Health Services Guirguis, L; University of Alberta, Faculty of Pharmacy; University of Alberta, Hayward, Kathleen; Alberta Health Services Lohmann, Tara; University of Calgary, Medicine MacLean, Joanna; University of Alberta, Pediatrics Manafo, Elizabeth; Independent Consultant Paskey, Janice; Alberta Health Services Rasiah, Jananee; University of Alberta Rimkus, Mark; Alberta Health Services Rizvi, Syeda ; Alberta Health Services Robinson, Gerry; Alberta Health Services Seefried, Brent; Alberta Health Services Somani, Zeeyaan; Alberta Health Services Tindall, Mindy; Alberta Health Services Vliagoftis, Harissios; University of Alberta Pendharkar, Sachin; University of Calgary, Medicine Stickland , Michael K ; University of Alberta,
<b>Primary Subject Heading</b>:	Respiratory medicine
Secondary Subject Heading:	Patient-centred medicine
Keywords:	Asthma < THORACIC MEDICINE, SLEEP MEDICINE, RESPIRATORY MEDICINE (see Thoracic Medicine)

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**Title**

What Are the Respiratory Health Research Priorities in Alberta Canada? A Stakeholder Consultation

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Abstract Word Count: 229

Manuscript Word Count: 3,327

Number of Figures: 0

Number of Tables: 3

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What Are the Respiratory Health Research Priorities in Alberta Canada? A Stakeholder Consultation

**ABSTRACT**

**Objective-** The Respiratory Health Strategic Clinical Network (RHSCN) was launched to facilitate respiratory and sleep health through implementation of innovative, patient-centred, evidence-informed coordinated services in Alberta. In collaboration with project partners, the RHSCN aimed to determine- the respiratory research priorities for Alberta. **Design-** The four phases of this research prioritization project included (1) identifying research questions from stakeholders, (2) determining which research questions had been answered in existing literature, (3) prioritizing unanswered questions, and (4) finalizing the priorities through an in-person workshop. **Setting-** The study occurred in Alberta, Canada, over a two-year period beginning in March 2017. **Participants-** A total of 446 patients, clinicians and other stakeholders consented to participate in the survey. **Results-** A total of 595 possible questions were submitted, with 343 unique questions identified. Of the questions, 95 were out of scope, 155 answered by existing literature, and ten were combined with others, while 83 were determined to be unanswered in the literature. Stakeholders were surveyed again to prioritize the remaining 83 questions, and they were reviewed by the project Steering Committee (clinicians and patients). At the in-person workshop, the Steering Committee identified 17 research topics as priority areas for respiratory and sleep research in Alberta. **Conclusion-** A stakeholder-led research prioritization process identified optimal clinical management/follow-up, equitable access to services, and management of social, psychological, mental health issues related to respiratory/sleep health as priority research areas.

## STRENGTHS AND LIMITATIONS OF THE STUDY

- Patients, caregivers, clinicians, and researchers identified the respiratory research priorities to assist in determining how research funding/support would be allocated, and to subsequently inform stakeholders of the chosen priorities.
- A four phase approach was used including (1) identifying research questions from stakeholders, (2) determining which research questions had been answered in existing literature, (3) prioritizing unanswered questions, and (4) finalizing the priorities through an in-person workshop.
- The RHSCN Research Prioritization process identified 17 priorities for respiratory and sleep health, integrating the perspectives of patients/caregivers and clinicians/researchers. Eight questions applied to both respiratory and sleep health, three were respiratory specific and six were sleep specific.
- These priorities included equitable access to health services, the economic impact of respiratory and sleep concerns, community-based management (such as referrals, self-management, when to seek medical care), and disease diagnosis, control and management.
- Key methodological limitations of the study include challenges associated with an online survey using convenience sampling such as readability and selection bias that may have impacted generalizability of study results. Additionally, we note respiratory research priorities may have shifted since the COVID-19 pandemic.

What Are the Respiratory Health Research Priorities in Alberta Canada? A Stakeholder Consultation

INTRODUCTION

In Alberta, respiratory disease is a prevalent, costly burden on the health care system. It is estimated that chronic obstructive pulmonary disease (COPD) costs the province over \$250 million per year[1], while asthma leads to missed days at school, frequent health care utilization, and negative impacts on families' quality of life.[2]–[4] Obstructive sleep apnea (OSA) is likely underdiagnosed in Canada[5] and may lead to reduced quality of life, workplace and vehicular accidents, and decreased work productivity[6]–[10]. In early 2014 the Respiratory Health Strategic Clinical Network (RHSCN) was launched by Alberta Health Services (the provincial health authority) to address the many challenges associated with respiratory and sleep disorders with the aim to facilitate optimal respiratory health through implementation of innovative, patient-centred, evidence-informed and coordinated services in the province of Alberta.[11] The RHSCN supports all respiratory-related conditions, with priority areas being asthma, COPD, and sleep-disordered breathing. The RHSCN consists of a Core Committee, which guides the overall direction of the Network, as well as several working groups that undertake specific projects to support the provision of evidence-based respiratory health care within the Alberta Health Services through quality improvement initiatives, development of clinical practice guidelines and implementation of innovative interventions. The scientific office within the RHSCN facilitates clinical research through funding and research support to promote clinical respiratory research, uptake of best evidence, building research capacity in the province and directs the Network's research priorities.

Historically, research agendas were dictated by the scientific community; however, there is a growing recognition that patients, caregivers, and clinicians should be engaged in identifying research priorities.[12] Increasingly in Canada there have been several research prioritization activities that used a patient-oriented approach.[13]–[15] The James Lind Alliance (JLA) suggests that patients, caregivers and clinicians work together to identify treatment uncertainties related to specific conditions and

prioritize those they collectively agree to be most important.[16] Using a modified JLA approach, the purpose of this project was to engage patient, caregiver, clinician, and researcher stakeholders to identify the respiratory research priorities that would assist the RHSCN scientific office in determining how research funding/support would be allocated, and to subsequently inform stakeholders of the chosen priorities.

## **METHODS**

### **Steering Committee Development**

The project was conducted from March 2017 to February 2019. Project oversight was performed by a Steering Committee. Patient engagement professionals from Alberta Health Services and Alberta Strategy for Patient-Oriented Research Support for People and Patient-Oriented Research and Trials Unit (AbSPORU) helped to identify patients/caregivers who might be interested in Steering Committee membership. Patients and caregivers met with the project lead (HS) by phone to discuss the scope of the project and required commitment. Steering Committee members were not compensated beyond travel expenses. The steering committee comprised of four individuals with personal experience, two caregivers, four respiratory physicians (broadly representing clinicians from paediatrics, sleep medicine, asthma/allergy and COPD), two respiratory therapists, two pharmacists, one nurse and the Scientific Director of the RHSCN; within the group there was representation from academic and community-based clinicians. We did not request Steering Committee members to disclose personal information related to dimensions of diversity, however the group acknowledged the importance of broad representation, felt the group was generally representative, while respecting the privacy of the team members.

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**Priority Setting Process**

The initial in-person meeting of the Steering Committee provided an opportunity to introduce the project, discuss examples of similar work from other groups,[14] and review the process of priority setting. As the RHSCN addresses both respiratory and sleep health concerns, both topics were included in the priority setting and deemed equally important.

The Steering Committee chose to follow a modified James Lind Alliance (JLA) Priority Setting Partnership method, as described by The Alberta Depression Research Priority Setting Project.[14] This approach was selected as it had demonstrated feasibility, rigor, and significant patient/caregiver engagement[17]. The adopted process was similar to the JLA method, however the voices of individuals with personal experience participated throughout (without the funnel approach, whereby the number of participants are reduced at each phase) with an egalitarian, consensus-building strategy within the Steering Committee [13]. The four-phase process included (1) identifying potential research questions by stakeholders, (2) determining research questions that had been answered, (3) prioritizing unanswered questions, and (4) finalizing the priority list at an in-person workshop.

**Participant Recruitment**

Study participants included individuals that participated in the online surveys designed to elicit their research priorities. We distributed the survey using posters, cards with QR codes and survey links (respiratory and sleep clinics in the province (both adult and paediatric), health care professional respiratory special interest groups, social media platforms, and patient advocacy and support groups). Participants were asked to share the survey link widely. The RHSCN further shared the survey links through provincial respiratory/sleep newsletters and email communications to members. The survey aimed to include three groups: (1) individuals with personal experience of respiratory disease or sleep disorders, (2) caregivers of individuals with respiratory disease or sleep disorders, and (3) health care

providers to individuals with respiratory disease or sleep disorders. Participants self-identified to which group(s) they belonged via the survey. Participants were not directly reimbursed but offered to enter a draw for a gift card prize for participation.

### **Patient and Public Involvement**

This project was co-led by patient/caregiver team members of the Steering Committee. Every phase of the methodology included patient/caregiver involvement, as they were equal members and active decision makers. Patients and caregivers were involved in the study design, implementation, data analysis and manuscript preparation (including authorship). Additionally, knowledge translation activities will include working with patient/family groups to disseminate the research prioritization questions.

### **Ethics Approval**

Ethics approval was received by the health research ethics board of the University of Alberta (Pro00075354).

### **DATA ANALYSIS**

#### **Phase 1: Identify Potential Research Questions by Stakeholders**

To obtain broad input of potential research questions, the Steering Committee designed an initial survey asking two open-ended questions:

1. What questions do you have about breathing problems?
2. What questions do you have about sleep problems?

Participants were provided with rationale and context for the survey, information about how the data would be used, contact information for seeking further information and asked to provide their consent (by clicking a consent box on the online survey) to participate and use their responses for the study.

They were invited to answer one or both questions. They were also asked optional questions including



gender and age group. The survey was designed to take less than ten minutes to complete and was first reviewed/tested by members of the Steering Committee with personal experience with respiratory health or sleep concerns. The survey was completed online over a two-month period (winter 2017) using the Research Electronic Data Capture (REDCap) online platform, hosted by the University of Calgary. REDCap is a secure, web-based software platform designed to support data capture for research studies.[18], [19] Incomplete surveys (those without submitted questions) were excluded from the analysis.

**Phase 2: Determine Research Questions that Had Been Answered**

The supplementary material provides a detailed summary of the process used to identify whether the submitted questions had already been answered in the literature. In brief, research questions that were submitted by participants via the survey in Phase 1 were reviewed by two members of the team. Duplicate questions were removed and similar questions were collated. Questions were reframed where necessary to ensure they identified the patient population, intervention, control and outcome (PICO) where possible. For each question topic, the following databases were searched for relevant, up-to-date systematic reviews, clinical practice guidelines and/or study protocols (i.e., studies in process) that addressed the specific research question:

- Cochrane Database of Systematic Reviews (EBM Reviews)
- American College of Physicians (ACP) Journal Club via Medline (Ovid)
- Database of Abstracts of Reviews of Effects (DARE) via Medline (Ovid)
- Medline databases (Ovid)
- Canadian Respiratory Guidelines (Canadian Thoracic Society)
- Global Initiative for Asthma (GINA)
- AAFP (USA)

- NICE guidelines (UK)

Research questions were defined as answered if we were able to provide a summary conclusion addressing the question and citation for the reference. Questions that were determined not to be adequately answered in the literature (i.e. the topic was not addressed by the above sources) were included in the list of unanswered questions to move forward to Phase 3, while questions confirmed to have been already answered did not move forward to Phase 3. Although policy and knowledge translation questions were initially considered out of scope, they were provided to the Steering Committee in a separate list for their consideration in Phase 3. This was done at the request of the Steering Committee as the group felt that knowledge translation and policy questions may have significant relevance to the work of the RHSCN.

### **Phase 3: Prioritization of Unanswered Questions**

The final list of unanswered questions was divided into two categories: 1) questions relating to breathing problems and 2) questions relating to sleep problems. Participants in Phase 1 were contacted again and asked to select their top ten priorities from one or both categories via a new REDCap survey (spring 2018). Data from this survey was used to inform the Steering Committee's discussion during the final workshop.

### **Phase Four: Final In-Person Priority Setting Workshop**

The Steering Committee participated in an in-person workshop with additional participation by health professional groups and patient/caregivers that were underrepresented, such as pharmacy and parents/guardians, with a total of 11 participants. The workshop was led by a trained facilitator with experience working with the Strategic Clinical Networks and an understanding of Alberta's health care system. The workshop included an overview of the results of the two surveys, and the RHSCN scope and

work. Following a discussion of the results, the Steering Committee participants split into two groups to prioritize the existing sleep research questions and respiratory research questions. The two groups then worked collectively to compare priority rankings, remove questions that were deemed as low priority by both groups. The Steering Committee mutually discussed the remaining questions and subsequently conducted a second round of prioritization as one group, again removing low priority questions. The final list of questions was determined through group consensus, and included combining some questions when appropriate and refining wording.

During the workshop, participants made the following key decisions to guide the work:

1. The list should not be guided by an arbitrary number (e.g., top ten); rather, the most important questions should be included in the list.
2. Several ‘out of scope’ questions were important and should not be dismissed without consideration, despite this approach being atypical from research prioritization methods
3. The significant overlap between respiratory and sleep health created redundancies in two lists, and a hybrid approach was developed.

**RESULTS**

We received 461 responses to the first survey (Table 1). Thirteen participants’ responses were removed as they did not clearly provide consent, leaving a total of 448 survey responses. The survey methods were successful at gaining broad representation from the three core groups: 317 participants stated they were an individual with personal experience with respiratory disease/patient (71.1%), 207 stated they were a caregiver of an individual with respiratory disease (46.4%) and 276 stated they were a health care professional that provided care for individuals with respiratory disease (61.9%). Of note, many respondents selected more than one category with which they identified. See Table 1 for participants’ characteristics.

A total of 595 research questions were submitted. Two data scientists reviewed the questions for redundancy, leaving 343 unique questions. Of the remaining questions, 66 were determined to be answered in the existing literature while 94 were judged to be out of scope (policy and cost related questions). Of the 183 research questions remaining, five questions were deemed not to be research questions, 86 were mapped to evidence and identified as answered, while 92 were identified as unanswered, and remained in the list of unanswered questions for consideration. The Steering Committee determined that of the 92 questions, three questions were answered in the literature, one question was determined to be out of scope, and five were combined with other questions or deemed not to be research questions, leaving 83 possible research questions. Table 2 provides an overview of the mapping of the final research questions and answered questions by category.

One hundred forty stakeholders participated in the second survey to rank the remaining questions. The in-person workshop allowed the Steering Committee to review the remaining questions collectively and identify 17 final priority areas for respiratory and sleep research in Alberta. The final list included six questions for sleep health, three questions for respiratory health, and eight questions that were applicable to both sleep and respiratory health (see Table 3).

## DISCUSSION

The RHSCN Research Prioritization process identified 17 priorities for respiratory and sleep health, integrating the perspectives of patients/caregivers and clinicians/researchers. These priorities included equitable access to health services, the economic impact of respiratory and sleep concerns, community-based management (such as referrals, self-management, when to seek medical care), and disease diagnosis, control and management (Table 3). This research is novel as it was co-owned and co-developed by patient/caregivers and clinicians/researchers, and is the first Canadian research

prioritization project that aimed to address both respiratory and sleep health. This prioritization will assist with decision making for the RHSCN related to allocation of resources and Network priorities.

Including individuals with personal experience as co-owners in research increases legitimacy and opportunity for innovation.[20] Additionally, researchers have moral and ethical responsibilities to ensure that the voice of patients and families are represented in research priorities.[21] A strength of this research is the high participation of patients and caregivers in the process, which is reflected in the nature of the research questions prioritized. As suggested by Breault and colleagues[14], the higher the level of participation from individuals with lived experiences (and their caregivers), the more closely a research prioritization list will reflect the true priorities of the community. The questions identified in this study had a strong focus on policy, equitable access to services, and the management of social, psychological, mental health issues related to respiratory/sleep health. Over two-thirds of survey respondents indicated they were patients, and almost half stated they were caregivers of someone with a respiratory or sleep problem (individuals could select more than one role).

Respiratory specific research prioritization has also increased in prominence. In the Netherlands, patient focus groups and questionnaires were used to identify research priorities for asthma and COPD.[22] Two significant priorities that arose were knowledge about the causes of the diseases and development of more effective medications, with little emphasis upon socio-political factors that impact health care. In contrast, our priorities for respiratory/sleep health research had a strong emphasis on social, economic and health care access issues. This variance may be the result of the autonomy of the Steering Committee, who made modifications to the proposed structure and inclusion criteria of the priorities by including out of scope policy and economic questions in the final review, or a reflection of shifting priorities over time, and the nature of the RHSCN being focused on health service delivery, rather than basic science research.

Other targeted respiratory research prioritization activities identified some priorities with similar themes to this research. A formal JLA on asthma was previously conducted in the United Kingdom . [23] Our recent work is consistent with their JLA which identified the importance of managing adolescent and young adult care as an important priority, as well as identifying effective self-management interventions. Additional research has identified nursing-related respiratory research priorities through modified Delphi approaches.[24] This work determined patient understanding of asthma control and the clinical and cost-effectiveness of respiratory nurse interventions as the most prominent research priorities. [24] While these priorities were nursing-specific, cost/economic impact was highlighted as an important priority in this work as well. A separate modified Delphi was conducted in critical care, adult pulmonary, and sleep conditions across 45 countries to identify nursing research priorities to inform research and advise funding agencies. [25] This extensive project resulted in research priorities focused in four broad areas including: communication, education, risk reduction and psychological support. [25] Similar to this previous work, our project identified psychological factors that impact respiratory disease/sleep disorders and self-management supports (such as education) as important priorities.

In 2015, the American Thoracic Society released a statement on the importance of sleep health, and included research priorities. [26] This document highlighted the importance of conducting research linking sleep quality and health outcomes. [26] This priority, developed by international sleep medicine leaders is consistent with the research priority identified by our process (What strategies can be used to promote sleep as an important contributor to health?). A JLA initiative was recently completed in Saskatchewan, Canada to assess the research priorities for sleep apnea. [27] Alignment with our work shows common priorities related to equitable access, follow-up care/re-evaluation, and additional therapies (beyond CPAP) for OSA treatment. [27] Recent work in Quebec Canada surveyed 148 COPD patients to develop a health and healthcare research agenda for COPD. This work identified that relief of breathlessness was a research and health care priority for patients. (20). While management of

breathlessness was not specifically identified within the current prioritization process, we did identify self-management and community supports/resources as a research priority (For individuals living with respiratory disease/sleep disorders, what are the most effective self-management interventions and/or community supports/resources?). Research prioritization activities have increased in recent years, and similarities with our work are demonstrated, however this research is important as it was developed by stakeholders from across the healthcare system with a focus on driving the local respiratory and sleep research agenda.

**LIMITATIONS**

There are limitations to consider related to this research. First, an online survey platform was convenient, cost-effective and timely, however it may have prevented some individuals from participating in the research. Second, our sampling approach may have resulted in a selection bias, as participants self-selected their participation.. Moreover, ... each individual will have their own motivation for participating, making it difficult to generalize study results to the population. We did not undertake specific approaches to target Indigenous populations and caregivers, those with a primary language other than English, nor those with limited internet access. It is important to note that the study sample was predominantly female, which may have influenced the prioritization process. This sampling methodology may limit the generalizability of this study. In future research, we would advocate for requesting ethnicity/race information from participants and using this information to guide purposeful recruitment of participants. As well, we would suggest developing a targeted strategy for ensuring representation from those without internet access, and ensuring key populations are included.

Third, the time commitment for participating as a Steering Committee member was substantial. Members attended two full-day in-person meetings and approximately six hours of telephone meetings, plus numerous emails/material review over the course of 18 months. The participation level of the patient/caregiver steering committee demonstrated strong commitment, however future groups may

consider additional strategies, such as role-sharing, to decrease volunteer fatigue. Also, these members were identified specifically for participation in this work, which may have resulted in selection bias.

Fourth, this research focused on the province of Alberta, and while the results may be broadly translatable, generalizability may be limited. However, prioritization activities driven by local stakeholders are important to providing research priorities of value to the population. Finally, given the data for this study was collected over 2017-2019, there may be substantial changes in respiratory research priorities in Alberta, particularly in consideration of the COVID-19 pandemic.

## CONCLUSION

By involving individuals with personal experience with respiratory disease, caregivers and health care professionals, a research priority list comprising six questions for sleep health, three questions for respiratory health, and eight questions that were applicable to both sleep and respiratory health was developed. Broadly, this research was novel as the research priorities demonstrated an increased focus on issues such as equitable access, economic impact and community disease management (including resource allocation and self-management supports). This research exercise will lead to a more focused distribution of research funds and other resources locally, and will inform other groups as they look to support research that is more relevant to patients.



**Acknowledgements:** Our sincere appreciation to the Respiratory Health Section of the Medicine Strategic Clinical Network Core Committee for their ongoing support of this project. In addition, we thank all of the participants in the surveys and the research prioritization process. This work was supported by an in-kind contribution by the Respiratory Health Strategic Clinical Network™ of Alberta Health Services and the Alberta SPOR SUPPORT Unit Patient Engagement Platform.

**Funding statement:** This research received no specific grant from any funding agency in the public, commercial or not-for-profit sectors.

**Conflict of Interest Statement:** Ms. Hayward received honoraria for speaking at continuing education activities for health care professionals from Astra Zeneca in the last year, unrelated to this project. Dr. Sachin Pendharkar discloses he has received advisory fees from Jazz Pharmaceuticals, Paladin Labs and the International Centre for Professional Development in Health and Medicine; received research funding from Canadian Institute for Health Research, Alberta Health Services, The Lung Association, and MITACS; additionally, he has participated in a study by Jazz Pharmaceuticals (site co-investigator with no personal stipend or salary). Dr. Heather Sharpe & Dr. Michael Stickland received research funding from the Alberta Boehringer Ingelheim Collaboration and Dr. Sharpe is a member of the Board of Directors of the Canadian Network of Respiratory Care. The remaining authors have no other disclosures.

**Availability of data and materials-** the datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

**Ethics approval-** granted from the Health Research Ethics Board at the University of Alberta.

**Contributors’ Statement**

\*As this work was co-owned and developed in collaboration with our patient/caregiver advisors, all members of the Steering Committee were responsible for study design and interpretation of results. All authors have given permission for the submission of the manuscript, and have agreed to be listed as a co-author.

Heather Sharpe –led the Steering Committee (SC) through the study design process, data collection and analysis. Was responsible for drafting the manuscript, editing and is corresponding author.

Lisa Cerato-facilitated the final workshop to the creation of the final prioritization list. Provided feedback on the manuscript.

Darlene Derech-a patient/caregiver advisor that was a member of the SC. Involved in the study design, data collection, analysis and final research prioritization. Was responsible for providing feedback on the manuscript.

Lisa Guirguis-a health professional/academic representative on the SC. Involved in the study design, data collection, analysis and final research prioritization. Was responsible for providing feedback on the manuscript.

Kathy Hayward- a health professional/academic representative on the SC. Involved in the study design, data collection, analysis and final research prioritization. Was responsible for providing feedback on the manuscript.

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7 Joanna E. MacLean- a health professional/academic representative on the SC. Involved in the study  
8 design, data collection, analysis and final research prioritization. Was responsible for providing feedback  
9 on the manuscript.  
10

11 Elizabeth Manafo-conducted the determination if submitted research questions were answered in the  
12 existing literature and responsible for the methodology and reporting. Provided feedback on the  
13 manuscript.  
14

15 Janice Paskey - a patient/caregiver advisor that was a member of the SC. Involved in the study design,  
16 data collection, analysis and final research prioritization. Was responsible for providing feedback on the  
17 manuscript.  
18

19 Sachin R. Pendharkar- a health professional/academic representative on the SC. Involved in the study  
20 design, data collection, analysis and final research prioritization. Was responsible for providing feedback  
21 on the manuscript.  
22

23 Jananee Rasiah- a health professional/academic representative on the SC. Involved in the study design,  
24 data collection, analysis and final research prioritization. Was responsible for providing feedback on the  
25 manuscript.  
26

27 Mark Rimkus- a health professional/academic representative on the SC. Involved in the study design,  
28 data collection, analysis and final research prioritization. Was responsible for providing feedback on the  
29 manuscript.  
30

31 Syeda Kinza Rizvi- a patient/caregiver advisor that was a member of the SC. Involved in the study design,  
32 data collection, analysis and final research prioritization. Was responsible for providing feedback on the  
33 manuscript.  
34

35 Gerry Robinson - a patient/caregiver advisor that was a member of the SC. Involved in the study design,  
36 data collection, analysis and final research prioritization. Was responsible for providing feedback on the  
37 manuscript.  
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39 Brent Seefried- a health professional/academic representative on the SC. Involved in the study design,  
40 data collection, analysis and final research prioritization. Was responsible for providing feedback on the  
41 manuscript.  
42

43 Zeeyaan Somani - a patient/caregiver advisor that was a member of the SC. Involved in the study design,  
44 data collection, analysis and final research prioritization. Was responsible for providing feedback on the  
45 manuscript.  
46

47 Mindy Tindall- a patient/caregiver advisor that was a member of the SC. Involved in the study design,  
48 data collection, analysis and final research prioritization. Was responsible for providing feedback on the  
49 manuscript.  
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Harissios Vliagoftis- a health professional/academic representative on the SC. Involved in the study design, data collection, analysis and final research prioritization. Was responsible for providing feedback on the manuscript.

Michael K Stickland- the Scientific Director of the RHSCN. Involved in the study design, data collection, analysis and final research prioritization. Was responsible for providing feedback on the manuscript.

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Table 1. Survey participant demographic characteristics

Characteristic	No (%) of total respondents (n=448)
<b>Do you prefer to answer questions related to breathing problems, sleep problems or both? (n=409)</b>	
Breathing problems	81 (19.8)
Sleep problems	123 (30.1)
Both (breathing and sleep problems)	205 (50.1)
<b>Category-may choose more than 1 (n=446)</b>	
Person with breathing problems	132 (29.6)
Person with sleep problems	185 (41.5)
Family/caregiver of person with breathing problems	90 (20.2)
Family/caregiver of person with sleep problems	117 (26.2)
Health care professional-breathing problems	148 (33.2)
Health care professional-sleep problems	128 (28.7)
Other (dentist, scientist, patient with co-morbidities)	11 (2.5)
<b>Gender (n=446)</b>	
Female	347 (77.8)
Male	86 (19.3)
Non-Binary	5 (1.1)
Prefer not to respond/Did not respond	8 (1.8)
<b>Age</b>	
≤17 years	1 (0.2)
18-29 years	44 (9.9)
30-49 years	116 (26.0)
50-79 years	197 (44.2)
≥80 years	4 (0.9)
Did not respond	4 (0.9)

Table 2. Submitted Research Questions by Topic Reviewed and Finalized by Steering Committee

Total	Total questions	Answered in Literature	Remaining Research questions
Asthma	33	20	13
Combustibles*	4	1	3
COPD	3	2	1
Devices	13	4	9
Diagnostic	7	4	3
General Medicine	14	11	3
ILD	9	4	5
Nasal	4	3	1
OSA	25	19	6
Sleep	35	17	18
Other	25	4	21
Cost and policy**	0	0	0
Total	172	89	83

\*combustibles included cigarette smoking, vaping and cannabis, as well as secondary effects from these products.

\*\*cost and policy questions were deemed out of scope and represented in the out of scope data.

Table 3. List of Final Research Questions

<b>Questions for Respiratory and Sleep</b>	
1.	What is the relationship between social, psychological, and mental health issues and respiratory disease/sleep disorders for the individual/the family?
2.	What is the economic impact of respiratory disease/sleep disorders for community and inpatient/urgent care in Alberta?
3.	For individuals with suspected respiratory disease/sleep disorders, when is it appropriate for a primary care provider to refer to a specialist, compared to continuing care, to ensure the best treatment/health outcomes? What are the educational needs of primary care providers to facilitate this?
4.	What model of care is most effective at transitioning patients and their families from paediatric to adult respiratory/sleep care?
5.	For individuals living with respiratory disease/sleep disorders, what are the most effective self-management interventions and/or community supports/resources?
6.	What strategies will improve equitable access to respiratory/sleep care for Albertans?
7.	What are the patient & families' priorities related to the treatment of their breathing/sleep problems?
8.	For individuals with respiratory/sleep problems, which interventions, resources, programs in the community will result in fewer specialty care, emergency department or hospital visits?
<b>Questions for Respiratory</b>	
1.	How can we improve access to pulmonary rehabilitation for individuals with respiratory disease?
2.	For individuals with respiratory disease, how do environmental factors (humidity, air pollution etc.) impact disease control/management in Alberta?
3.	How does an individual with respiratory disease determine if different therapies will be worth the additional cost (money, risk of side effects, exacerbations)?
<b>Questions for Sleep</b>	
1.	For individuals with suspected sleep-related breathing disorders (e.g. obstructive sleep apnea), what is the recommended ongoing clinical management/follow-up care to improve and sustain health outcomes?
2.	Do treatments besides CPAP improve outcomes for individuals with sleep-related breathing disorders (e.g. obstructive sleep apnea and/or hypoventilation)?
3.	What is the current quality of provision and interpretation of investigations for sleep-related breathing disorders in Alberta?
4.	What strategies can be used to promote sleep as an important contributor to health?
5.	For individuals experiencing poor quality sleep, at what point should they seek medical advice to improve their health outcomes?
6.	What are the policy factors that inform insurable coverage for testing and treatment of sleep-related breathing disorders in Alberta?

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Appendix A. Verification Process to Identify Uncertainties

Where, no up-to-date, reliable systematic reviews or guidelines of research evidence addressing the uncertainty exist/have not been adequately addressed. If a study protocol exists, this will be noted. These uncertainties will move forward for interim prioritization.

Process	Description	Output
a. Check evidence-base for relevant, up to date systematic reviews, guidelines and study protocols (i.e., studies in process)	i. Cochrane Database of Systematic Reviews (EBM Reviews) ii. American College of Physicians (ACP) Journal Club via Medline (Ovid) iii. Database of Abstracts of Reviews of Effects (DARE) via Medline (Ovid) iv. Medline databases (Ovid) v. <a href="#">Canadian Respiratory Guidelines</a> (Canadian Thoracic Society) vi. <a href="#">Global Initiative for Asthma</a> (GINA) vii. <a href="#">AAFP</a> (USA) viii. <a href="#">NICE guidelines</a> (UK) ix. Other (used if unique population, intervention or outcome would be better addressed in a different database)	If evidence is available, cite it with primary author conclusion.
	Using specific search terms as they relate to the submission topic area, subtopic area and outcome (as identified). Specific search outputs will be documented as they relate to each topic and subtopic area.	
	To check questions about sleep and breathing risk factors, which are mainly addressed by reviews including prospective cohort studies, other systematic reviews. MEDLINE, EMBASE, CINAHL and PsycINFO will be searched with the same keywords using a systematic review filter.	
b. Determine if evidence is reliable and relevant using	1] Systematic review	
	1.1 Has it been published in the last 3 years?	[YES > does not move forward; NO > moves forward]

Process	Description	Output
the following criteria:	<p><b>AND</b></p> <p>1.2 For evidence not derived from Cochrane et al., ask the following: Does it have published clear methods?</p> <ul style="list-style-type: none"> <li>• Where inclusion/exclusion criteria reported?</li> <li>• Was search adequate?</li> <li>• Where the included studies synthesized?</li> <li>• Was the quality of included studies assessed?</li> <li>• Are sufficient details about the individual included studies presented?</li> <li>• Was any potential bias addressed/managed?</li> </ul>	[YES > does not move forward; NO > moves forward]
	<p><b>AND</b></p> <p>1.3 Are reported findings clinically significant?</p>	[YES > does not move forward; NO > moves forward]
	<b>2] Guidelines</b>	
	2.1 Are guidelines evidence-based, that are used as a framework for clinical decision making and supporting best practices from authoritative Canadian, American or UK bodies?	[YES > does not move forward; NO > moves forward]
	<b>3] Protocol</b>	
	3.1 If no systematic review or guidelines have been located, conduct Protocol search through EBM Cochrane	[YES > note its existence; NO > moves forward]
		<p>If <b>all of the criteria</b> have been answered with a <b>Yes = "Known unknown"</b> [tier 2]</p> <p>If <b>one or more</b> of the criteria has been answered with a <b>No = Uncertainty</b></p>

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Process	Description	Output
c. Verify selection output	Clinical significance must be determined by Steering Committee based on reported findings and recorded author conclusions.	Long-list of uncertainties are identified for interim prioritization (and 'known unknowns' are recorded with evidence cited)

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### Reporting guideline for health research priority setting with stakeholders (REPRISE)

No	Item	Descriptor and/or examples
A	Context and scope	
1	Define geographical scope	Provincial scope
2	Define health area, field, focus	Respiratory and sleep health
3	Define the intended beneficiaries	Individuals living with respiratory and sleep disorders in the province of Alberta and their families.
4	Define the target audience of the priorities	Respiratory Health Section of the Medicine Strategic Clinical Network-to use the prioritization to fund research
5	Identify the research area	Health service research and clinical research
6	Identify the type of research questions	Was not pre-defined
7	Define the time frame	To provide research prioritization for five years
B	Governance and team	
8	Describe the selection and structure of the leadership and management team	The Scientific Office of the Network was primarily responsible for the development of the Steering Committee. The Steering Committee was created to ensure a strong voice for those with lived experience of respiratory and/or sleep disorders. The Core Committee of the Network provided content expertise.
9	Describe the characteristics of the team	Project oversight was performed by a Steering Committee comprising four individuals with lived experience, two caregivers, four respiratory physicians,

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No	Item	Descriptor and/or examples
		two respiratory therapists, two pharmacists, one nurse and the Scientific Director of the RHSCN, within the group there was representation from academic and community based clinicians. We did not request Steering Committee members to disclose sex/gender or race/ethnicity, however the group was diverse in perceived age, sex, gender, ethnicity and academic/non-academic background.
10	Describe any training or experience relevant to conducting priority setting	The Patient Engagement Platform of the Alberta Strategy for Patient-Oriented Research's Support for People and Patient-Oriented Research and Trials Unit provided guidance, a consultant with priority setting experience was hired to assist with identifying unanswered questions and a facilitator was brought to the team for the final priority setting workshop.
C	Framework for priority setting	
11	State the framework used (if any)	A modified James Lind Alliance approach was used based upon the Alberta Depression Research Priority Setting Project
D	Stakeholders or participants	
12	Define the inclusion criteria for stakeholders involved in priority-setting	Patients, caregivers, health professionals, researchers, policy makers, and other stakeholders that self-identified as having an interest in respiratory/sleep health
13	State the strategy or method for identifying and engaging stakeholders	Posters and cards with QR codes and website links were disseminated through stakeholders, such as respiratory and sleep clinics. Steering Committee members were asked to share the flyer within their community (such as support groups, online forums etc.) and through social media platforms. The

No	Item	Descriptor and/or examples
		RHSCN shared the survey links through newsletters and email communications to members.
14	Indicate the number of participants and/or organizations involved	A total of 461 unique individuals accessed the first survey. Fifteen participants' responses were removed as they did not provide clear consent, leaving a total of 446 survey responses.
15	Describe the characteristics of stakeholders	Three core groups: 317 participants stated they were an individual with lived experience with respiratory disease/patient (71.1%), 207 stated they were a caregiver of an individual with respiratory disease (46.4%) and 276 stated they were a health care professional that provided care for individuals with respiratory disease (61.9%). Of note, many respondents selected more than one category with which they identified.
16	State if reimbursement for participation was provided	Individuals that participated on the Steering Committee were provided reimbursement for any expenses associated with attending the in-person meetings including travel (hotel, mileage, parking, meals etc.). Participants were not paid an honoraria or other compensation. Participants in the survey were invited to enroll in a draw for a small gift card to a book store for participating.
E	Identification and collection of research priorities	
17	Describe methods for collecting initial priorities	The four phases to the project included (1) identifying research questions by stakeholders, (2) determining research questions that had been answered in existing literature (guidelines, systematic reviews etc.), (3) prioritization of unanswered questions, and (4) finalization through an in-person priority setting workshop. The Steering Committee designed the initial survey asking



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No	Item	Descriptor and/or examples
		two open-ended core questions, (1) What questions do you have about breathing problems? And (2) What questions do you have about sleep problems?
18	Describe methods for collating and categorizing priorities	Priorities were originally organized into the broad categories of respiratory health and sleep health. Subsequently they were categorized into sub-groups to facilitate data management. For example, respiratory health priorities were organized into: asthma, COPD, medications, devices, obstructive sleep apnea, cost & policy, diagnostic, nasal, sleep, interstitial lung disease, and other.
19	Describe methods and reasons for modifying (removing, adding, reframing) priorities	All proposed priorities were reframed as necessary to create a PICO (patient, intervention, control & outcome) question were possible. Priorities that overlapped were combined.
20	Describe methods for refining or translating priorities into research topics or questions	Two members of the team were responsible for creating research questions from the submissions. The resulting list was reviewed by the Steering Committee.
21	Describe methods for checking whether research questions or topics have been answered	For each question topic, key databases were searched for relevant, up-to-date systematic reviews, clinical practice guidelines and/or study protocols (i.e., studies in process). Questions that had not been adequately answered in the literature (the topic was not addressed by the above sources), were to be included in the list of unanswered questions. Questions that were answered in the literature were identified as 'known unknowns', and along with possibly out of scope questions (policy questions or those that would not be answered by the literature sources) were included by the two reviewers in a separate list for review by list to the Steering Committee. Questions for final inclusion

No	Item	Descriptor and/or examples
		based on clinical and research significance were determined by the Steering committee.
22	Describe number of research questions or topics	A total of 595 research questions were submitted (see Figure 1). A team of two people identified answers for 343 (74%) questions within the literature. A total of 178 research questions were identified as unanswered questions that moved forward. One hundred forty stakeholders participated in the second survey to rank the remaining questions. A final list of 17 research topics was identified as priority areas for respiratory and sleep research in Alberta through this prioritization.
F	Prioritization of research topics/questions	
23	Describe methods and criteria for prioritizing research topics or questions	Two online surveys were completed. The first, to identify possible priorities, the second to prioritize the identified priorities from the first survey. The Network Core Committee reviewed the results and provided their input to the Steering Committee. An in person priority setting workshop was completed to identify the final priorities.
24	State the method or threshold for excluding research topics/questions	A consensus approach was used. The Steering Committee reached consensus on what questions would be included in the final list.
G	Output	
25	State the approach to formulating the research priorities	PICO (population, intervention, comparator, outcome) questions were formulated, however the Steering Committee did revise the priorities in the final priority setting workshop.
H	Evaluation and feedback	

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No	Item	Descriptor and/or examples
26	Describe how the process of prioritization was evaluated	The prioritization process has not yet been formally evaluated, however the impact will be assessed through the Scientific Office of the Network.
27	Describe how priorities were fed back to stakeholders and/or to the public; and how feedback (if received) was addressed and integrated	The results of the priority setting were shared with the RHSCN, including the Core Committee and the working groups.
I	Implementation	
28	Outline the strategy or action plans for implementing priorities	The priorities have been used to guide the funding of research grants offered by the Network, including seed grant funding and studentships.
29	Describe plans, strategies, or suggestions to evaluate impact	The Network evaluates the impact of the scientific office annually, as well as is involved in the evaluation and priority setting for the Network every 3-5 years.
J	Funding and conflict of interest	
30	State sources of funding	This work was supported by an in-kind contribution by the Respiratory Health Strategic Clinical Network™ of Alberta Health Services and the Alberta SPOR SUPPORT Unit Patient Engagement Platform.
31	Declare any conflicts or competing interests	The authors have no other disclosures.

### Checklist for Reporting Results of Internet E-Surveys (CHERRIES)

<b>Checklist Item</b>	<b>Explanation</b>	<b>Page Number</b>
Describe survey design	Describe target population, sample frame. Is the sample a convenience sample? (In “open” surveys this is most likely.)	Page 6-7
IRB approval	Mention whether the study has been approved by an IRB.	Page 7
Informed consent	Describe the informed consent process. Where were the participants told the length of time of the survey, which data were stored and where and for how long, who the investigator was, and the purpose of the study?	Page 7
Data protection	If any personal information was collected or stored, describe what mechanisms were used to protect unauthorized access.	n/a
Development and testing	State how the survey was developed, including whether the usability and technical functionality of the electronic questionnaire had been tested before fielding the questionnaire.	Page 7
Open survey versus closed survey	An “open survey” is a survey open for each visitor of a site, while a closed survey is only open to a sample which the investigator knows (password-protected survey).	Page 7
Contact mode	Indicate whether or not the initial contact with the potential participants was made on the Internet. (Investigators may also send out questionnaires by mail and allow for Web-based data entry.)	Page 7
Advertising the survey	How/where was the survey announced or advertised? Some examples are offline media (newspapers), or online (mailing lists – If yes, which ones?) or banner ads (Where were these banner ads posted and what did they look like?). It is important to know the wording of the announcement as it will heavily influence who chooses to participate. Ideally the survey announcement should be published as an appendix.	Page 6
Web/E-mail	State the type of e-survey (eg, one posted on a Web site, or one sent out through e-mail). If it is an e-mail survey, were the responses entered manually into a database, or was there an automatic method for capturing responses?	Page 6
Context	Describe the Web site (for mailing list/newsgroup) in which the survey was posted. What is the Web site about, who is visiting it, what are visitors normally looking for? Discuss to what degree the content of the Web site could pre-select the sample or influence the results. For example, a survey about vaccination on a anti-immunization Web site will have different results from a Web survey conducted on a government Web site	Page 6
Mandatory/voluntary	Was it a mandatory survey to be filled in by every visitor who wanted to enter the Web site, or was it a voluntary survey?	Page 6
Incentives	Were any incentives offered (eg, monetary, prizes, or non-monetary incentives such as an offer to provide the survey results)?	Page 6

Time/Date	In what timeframe were the data collected?	Page 6
Randomization of items or questionnaires	To prevent biases items can be randomized or alternated.	n/a
Adaptive questioning	Use adaptive questioning (certain items, or only conditionally displayed based on responses to other items) to reduce number and complexity of the questions.	n/a
Number of Items	What was the number of questionnaire items per page? The number of items is an important factor for the completion rate.	Page 7
Number of screens (pages)	Over how many pages was the questionnaire distributed? The number of items is an important factor for the completion rate.	Page 7
Completeness check	It is technically possible to do consistency or completeness checks before the questionnaire is submitted. Was this done, and if “yes”, how (usually JavaScript)? An alternative is to check for completeness after the questionnaire has been submitted (and highlight mandatory items). If this has been done, it should be reported. All items should provide a non-response option such as “not applicable” or “rather not say”, and selection of one response option should be enforced.	n/a
Review step	State whether respondents were able to review and change their answers (eg, through a Back button or a Review step which displays a summary of the responses and asks the respondents if they are correct).	n/a
Unique site visitor	If you provide view rates or participation rates, you need to define how you determined a unique visitor. There are different techniques available, based on IP addresses or cookies or both.	n/a
View rate (Ratio of unique survey visitors/unique site visitors)	Requires counting unique visitors to the first page of the survey, divided by the number of unique site visitors (not page views!). It is not unusual to have view rates of less than 0.1 % if the survey is voluntary.	n/a
Participation rate (Ratio of unique visitors who agreed to participate/unique first survey page visitors)	Count the unique number of people who filled in the first survey page (or agreed to participate, for example by checking a checkbox), divided by visitors who visit the first page of the survey (or the informed consents page, if present). This can also be called “recruitment” rate.	n/a
Completion rate (Ratio of users who finished the survey/users who	The number of people submitting the last questionnaire page, divided by the number of people who agreed to participate (or submitted the first survey page). This is only relevant if there is a separate “informed consent” page or if the survey goes over several pages. This is a measure for attrition. Note that	n/a

agreed to participate)	“completion” can involve leaving questionnaire items blank. This is not a measure for how completely questionnaires were filled in. (If you need a measure for this, use the word “completeness rate”.)	
Cookies used	Indicate whether cookies were used to assign a unique user identifier to each client computer. If so, mention the page on which the cookie was set and read, and how long the cookie was valid. Were duplicate entries avoided by preventing users access to the survey twice; or were duplicate database entries having the same user ID eliminated before analysis? In the latter case, which entries were kept for analysis (eg, the first entry or the most recent)?	n/a
IP check	Indicate whether the IP address of the client computer was used to identify potential duplicate entries from the same user. If so, mention the period of time for which no two entries from the same IP address were allowed (eg, 24 hours). Were duplicate entries avoided by preventing users with the same IP address access to the survey twice; or were duplicate database entries having the same IP address within a given period of time eliminated before analysis? If the latter, which entries were kept for analysis (eg, the first entry or the most recent)?	n/a
Log file analysis	Indicate whether other techniques to analyze the log file for identification of multiple entries were used. If so, please describe.	n/a
Registration	In “closed” (non-open) surveys, users need to login first and it is easier to prevent duplicate entries from the same user. Describe how this was done. For example, was the survey never displayed a second time once the user had filled it in, or was the username stored together with the survey results and later eliminated? If the latter, which entries were kept for analysis (eg, the first entry or the most recent)?	n/a
Handling of incomplete questionnaires	Were only completed questionnaires analyzed? Were questionnaires which terminated early (where, for example, users did not go through all questionnaire pages) also analyzed?	Page 8
Questionnaires submitted with an atypical timestamp	Some investigators may measure the time people needed to fill in a questionnaire and exclude questionnaires that were submitted too soon. Specify the timeframe that was used as a cut-off point, and describe how this point was determined.	n/a
Statistical correction	Indicate whether any methods such as weighting of items or propensity scores have been used to adjust for the non-representative sample; if so, please describe the methods.	n/a

This checklist has been modified from Eysenbach G. Improving the quality of Web surveys: the Checklist for Reporting Results of Internet E-Surveys (CHERRIES). J Med Internet Res. 2004 Sep 29;6(3):e34 [erratum in J Med Internet Res. 2012; 14(1): e8.]. Article available at <https://www.jmir.org/2004/3/e34/>; erratum available <https://www.jmir.org/2012/1/e8/>. Copyright ©Gunther Eysenbach. Originally published in the [Journal of Medical Internet Research](https://www.jmir.org/2004/3/e34/), 29.9.2004 and 04.01.2012.

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# BMJ Open

## What Are the Respiratory Health Research Priorities in Alberta Canada? A Stakeholder Consultation

Journal:	<i>BMJ Open</i>
Manuscript ID	bmjopen-2021-059326.R2
Article Type:	Original research
Date Submitted by the Author:	30-May-2022
Complete List of Authors:	Sharpe, Heather; University of Alberta, Medicine Cerato, Lisa; Alberta Health Services Derech, Darlene; Alberta Health Services Guirguis, L; University of Alberta, Faculty of Pharmacy; University of Alberta, Hayward, Kathleen; Alberta Health Services Lohmann, Tara; University of Calgary, Medicine MacLean, Joanna; University of Alberta, Pediatrics Manafo, Elizabeth; Independent Consultant Paskey, Janice; Alberta Health Services Rasiah, Jananee; University of Alberta Rimkus, Mark; Alberta Health Services Rizvi, Syeda ; Alberta Health Services Robinson, Gerry; Alberta Health Services Seefried, Brent; Alberta Health Services Somani, Zeeyaan; Alberta Health Services Tindall, Mindy; Alberta Health Services Vliagoftis, Harissios; University of Alberta Pendharkar, Sachin; University of Calgary, Medicine Stickland , Michael K ; University of Alberta,
<b>Primary Subject Heading</b>:	Respiratory medicine
Secondary Subject Heading:	Patient-centred medicine
Keywords:	Asthma < THORACIC MEDICINE, SLEEP MEDICINE, RESPIRATORY MEDICINE (see Thoracic Medicine)

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**Title**

What Are the Respiratory Health Research Priorities in Alberta Canada? A Stakeholder Consultation

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Abstract Word Count: 229

Manuscript Word Count: 3,327

Number of Figures: 0

Number of Tables: 3

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What Are the Respiratory Health Research Priorities in Alberta Canada? A Stakeholder Consultation

**ABSTRACT**

**Objective-** The Respiratory Health Strategic Clinical Network (RHSCN) was launched to facilitate respiratory and sleep health through implementation of innovative, patient-centred, evidence-informed coordinated services in Alberta. In collaboration with project partners, the RHSCN aimed to determine- the respiratory research priorities for Alberta. **Design-** The four phases of this research prioritization project included (1) identifying research questions from stakeholders, (2) determining which research questions had been answered in existing literature, (3) prioritizing unanswered questions, and (4) finalizing the priorities through an in-person workshop. **Setting-** The study occurred in Alberta, Canada, over a two-year period beginning in March 2017. **Participants-** A total of 446 patients, clinicians and other stakeholders consented to participate in the survey. **Results-** A total of 595 possible questions were submitted, with 343 unique questions identified. Of the questions, 95 were out of scope, 155 answered by existing literature, and ten were combined with others, while 83 were determined to be unanswered in the literature. Stakeholders were surveyed again to prioritize the remaining 83 questions, and they were reviewed by the project Steering Committee (clinicians and patients). At the in-person workshop, the Steering Committee identified 17 research topics as priority areas for respiratory and sleep research in Alberta. **Conclusion-** A stakeholder-led research prioritization process identified optimal clinical management/follow-up, equitable access to services, and management of social, psychological, mental health issues related to respiratory/sleep health as priority research areas.

## STRENGTHS AND LIMITATIONS OF THE STUDY

- Patients, caregivers, clinicians, and researchers identified the respiratory and sleep health research priorities to assist in determining how research funding/support would be allocated, and to subsequently inform stakeholders of the chosen priorities.
- A four phase approach was used including (1) identifying research questions from stakeholders, (2) determining which research questions had been answered in existing literature, (3) prioritizing unanswered questions, and (4) finalizing the priorities through an in-person workshop.
- Methodological limitations of the study include: challenges associated with an online survey such as readability, using convenience sampling, the possibility of selection and motivation bias, and the inability to measure sampling error, all of which may have impacted generalizability of study results.
- Additionally, we anticipate that respiratory research priorities may have shifted since the COVID-19 pandemic.

What Are the Respiratory Health Research Priorities in Alberta Canada? A Stakeholder Consultation

INTRODUCTION

In Alberta, respiratory disease is a prevalent, costly burden on the health care system. It is estimated that chronic obstructive pulmonary disease (COPD) costs the province over \$250 million per year[1], while asthma leads to missed days at school, frequent health care utilization, and negative impacts on quality of life.[2]–[4] Obstructive sleep apnea (OSA) is likely underdiagnosed in Canada[5] and may lead to reduced quality of life, workplace and vehicular accidents, and decreased work productivity[6]–[10]. In early 2014, the Respiratory Health Strategic Clinical Network (RHSCN) was launched by Alberta Health Services (the provincial health authority) to address the many challenges associated with respiratory and sleep disorders with the aim to facilitate optimal respiratory health through implementation of innovative, patient-centred, evidence-informed and coordinated services in the province of Alberta.[11] The RHSCN supports all respiratory-related conditions, with priority areas being asthma, COPD, and sleep-disordered breathing. The RHSCN consists of a Core Committee, which guides the overall direction of the Network, as well as several working groups that undertake specific projects to support the provision of evidence-based respiratory health care within the Alberta Health Services through quality improvement initiatives, development of clinical practice guidelines and implementation of innovative interventions. The scientific office within the RHSCN facilitates clinical research through funding and research support to promote clinical respiratory research, uptake of best evidence, building research capacity in the province and directs the Network’s research priorities.

Historically, research agendas were dictated by the scientific community; however, there is a growing recognition that patients, caregivers, and clinicians should be engaged in identifying research priorities.[12] Increasingly in Canada there have been several research prioritization activities that used a patient-oriented approach.[13]–[15] The James Lind Alliance (JLA) suggests that patients, caregivers and clinicians work together to identify treatment uncertainties related to specific conditions and

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2  
3 prioritize those they collectively agree to be most important.[16] Using a modified JLA approach, the  
4  
5 purpose of this project was to engage patient, caregiver, clinician, and researcher stakeholders to  
6  
7 identify the respiratory research priorities that would assist the RHSCN scientific office in determining  
8  
9 how research funding/support would be allocated, and to subsequently inform stakeholders of the  
10  
11 chosen priorities.  
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## 14 15 **METHODS**

### 16 17 **Steering Committee Development**

18  
19 The project was conducted from March 2017 to February 2019. Project oversight was performed by a  
20  
21 Steering Committee. Patient engagement professionals from Alberta Health Services and Alberta  
22  
23 Strategy for Patient-Oriented Research Support for People and Patient-Oriented Research and Trials  
24  
25 Unit (AbSPORU) helped to identify patients/caregivers who might be interested in Steering Committee  
26  
27 membership. Patients and caregivers met with the project lead (HS) by phone to discuss the scope of  
28  
29 the project and required commitment. Steering Committee members were not compensated beyond  
30  
31 travel expenses. The steering committee comprised of four individuals with personal experience, two  
32  
33 caregivers, four respiratory physicians (broadly representing clinicians from paediatrics, sleep medicine,  
34  
35 asthma/allergy and COPD), two respiratory therapists, two pharmacists, one nurse and the Scientific  
36  
37 Director of the RHSCN; within the group there was representation from academic and community-based  
38  
39 clinicians. We did not request Steering Committee members to disclose personal information related to  
40  
41 dimensions of diversity, however the group acknowledged the importance of broad representation, felt  
42  
43 the group was generally representative, while respecting the privacy of the team members.  
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**Priority Setting Process**

The initial in-person meeting of the Steering Committee provided an opportunity to introduce the project, discuss examples of similar work from other groups,[14] and review the process of priority setting. As the RHSCN addresses both respiratory and sleep health concerns, both topics were included in the priority setting and deemed equally important.

The Steering Committee chose to follow a modified James Lind Alliance (JLA) Priority Setting Partnership method, as described by The Alberta Depression Research Priority Setting Project.[14] This approach was selected as it had demonstrated feasibility, rigor, and significant patient/caregiver engagement[17]. The adopted process was similar to the JLA method, however the voices of individuals with personal experience participated throughout (without the funnel approach, whereby the number of participants are reduced at each phase) with an egalitarian, consensus-building strategy within the Steering Committee [13]. The four-phase process included (1) identifying potential research questions by stakeholders, (2) determining research questions that had been answered, (3) prioritizing unanswered questions, and (4) finalizing the priority list at an in-person workshop.

**Participant Recruitment**

Study participants included individuals that participated in the online surveys designed to elicit their research priorities. We distributed the survey using posters, cards with QR codes and survey links (respiratory and sleep clinics in the province (both adult and paediatric), health care professional respiratory special interest groups, social media platforms, and patient advocacy and support groups). Participants were asked to share the survey link widely. The RHSCN further shared the survey links through provincial respiratory/sleep newsletters and email communications to members. The survey aimed to include three groups: (1) individuals with personal experience of respiratory disease or sleep disorders, (2) caregivers of individuals with respiratory disease or sleep disorders, and (3) health care

providers to individuals with respiratory disease or sleep disorders. Participants self-identified to which group(s) they belonged via the survey. Participants were not directly reimbursed but offered to enter a draw for a gift card prize for participation.

### **Patient and Public Involvement**

This project was co-led by patient/caregiver team members of the Steering Committee. Every phase of the methodology included patient/caregiver involvement, as they were equal members and active decision makers. Patients and caregivers were involved in the study design, implementation, data analysis and manuscript preparation (including authorship). Additionally, knowledge translation activities will include working with patient/family groups to disseminate the research prioritization questions.

### **Ethics Approval**

Ethics approval was received by the health research ethics board of the University of Alberta (Pro00075354).

### **DATA ANALYSIS**

#### **Phase 1: Identify Potential Research Questions by Stakeholders**

To obtain broad input of potential research questions, the Steering Committee designed an initial survey asking two open-ended questions:

1. What questions do you have about breathing problems?
2. What questions do you have about sleep problems?

Participants were provided with rationale and context for the survey, information about how the data would be used, contact information for seeking further information and asked to provide their consent (by clicking a consent box on the online survey) to participate and use their responses for the study.

They were invited to answer one or both questions. They were also asked optional questions including



gender and age group. The survey was designed to take less than ten minutes to complete and was first reviewed/tested by members of the Steering Committee with personal experience with respiratory health or sleep concerns. The survey was completed online over a two-month period (winter 2017) using the Research Electronic Data Capture (REDCap) online platform, hosted by the University of Calgary. REDCap is a secure, web-based software platform designed to support data capture for research studies.[18], [19] Incomplete surveys (those without submitted questions) were excluded from the analysis.

**Phase 2: Determine Research Questions that Had Been Answered**

The supplementary material provides a detailed summary of the process used to identify whether the submitted questions had already been answered in the literature. In brief, research questions that were submitted by participants via the survey in Phase 1 were reviewed by two members of the team. Duplicate questions were removed and similar questions were collated. Questions were reframed where necessary to ensure they identified the patient population, intervention, control and outcome (PICO) where possible. For each question topic, the following databases were searched for relevant, up-to-date systematic reviews, clinical practice guidelines and/or study protocols (i.e., studies in process) that addressed the specific research question:

- Cochrane Database of Systematic Reviews (EBM Reviews)
- American College of Physicians (ACP) Journal Club via Medline (Ovid)
- Database of Abstracts of Reviews of Effects (DARE) via Medline (Ovid)
- Medline databases (Ovid)
- Canadian Respiratory Guidelines (Canadian Thoracic Society)
- Global Initiative for Asthma (GINA)
- AAFP (USA)

- NICE guidelines (UK)

Research questions were defined as answered if we were able to provide a summary conclusion addressing the question and citation for the reference. Questions that were determined not to be adequately answered in the literature (i.e. the topic was not addressed by the above sources) were included in the list of unanswered questions to move forward to Phase 3, while questions confirmed to have been already answered did not move forward to Phase 3. Although policy and knowledge translation questions were initially considered out of scope, they were provided to the Steering Committee in a separate list for their consideration in Phase 3. This was done at the request of the Steering Committee as the group felt that knowledge translation and policy questions may have significant relevance to the work of the RHSCN.

### **Phase 3: Prioritization of Unanswered Questions**

The final list of unanswered questions was divided into two categories: 1) questions relating to breathing problems and 2) questions relating to sleep problems. Participants in Phase 1 were contacted again and asked to select their top ten priorities from one or both categories via a new REDCap survey (spring 2018). Data from this survey was used to inform the Steering Committee's discussion during the final workshop.

### **Phase Four: Final In-Person Priority Setting Workshop**

The Steering Committee participated in an in-person workshop with additional participation by health professional groups and patient/caregivers that were underrepresented, such as pharmacy and parents/guardians, with a total of 11 participants. The workshop was led by a trained facilitator with experience working with the Strategic Clinical Networks and an understanding of Alberta's health care system. The workshop included an overview of the results of the two surveys, and the RHSCN scope and

work. Following a discussion of the results, the Steering Committee participants split into two groups to prioritize the existing sleep research questions and respiratory research questions. The two groups then worked collectively to compare priority rankings, remove questions that were deemed as low priority by both groups. The Steering Committee mutually discussed the remaining questions and subsequently conducted a second round of prioritization as one group, again removing low priority questions. The final list of questions was determined through group consensus, and included combining some questions when appropriate and refining wording.

During the workshop, participants made the following key decisions to guide the work:

1. The list should not be guided by an arbitrary number (e.g., top ten); rather, the most important questions should be included in the list.
2. Several ‘out of scope’ questions were important and should not be dismissed without consideration, despite this approach being atypical from research prioritization methods
3. The significant overlap between respiratory and sleep health created redundancies in two lists, and a hybrid approach was developed.

**RESULTS**

We received 461 responses to the first survey (Table 1). Thirteen participants’ responses were removed as they did not clearly provide consent, leaving a total of 448 survey responses. The survey methods were successful at gaining broad representation from the three core groups: 317 participants stated they were an individual with personal experience with respiratory disease/patient (71.1%), 207 stated they were a caregiver of an individual with respiratory disease (46.4%) and 276 stated they were a health care professional that provided care for individuals with respiratory disease (61.9%). Of note, many respondents selected more than one category with which they identified. See Table 1 for participants’ characteristics.

A total of 595 research questions were submitted. Two data scientists reviewed the questions for redundancy, leaving 343 unique questions. Of the remaining questions, 66 were determined to be answered in the existing literature while 94 were judged to be out of scope (policy and cost related questions). Of the 183 research questions remaining, five questions were deemed not to be research questions, 86 were mapped to evidence and identified as answered, while 92 were identified as unanswered, and remained in the list of unanswered questions for consideration. The Steering Committee determined that of the 92 questions, three questions were answered in the literature, one question was determined to be out of scope, and five were combined with other questions or deemed not to be research questions, leaving 83 possible research questions. Table 2 provides an overview of the mapping of the final research questions and answered questions by category.

One hundred forty stakeholders participated in the second survey to rank the remaining questions. The in-person workshop allowed the Steering Committee to review the remaining questions collectively and identify 17 final priority areas for respiratory and sleep research in Alberta. The final list included six questions for sleep health, three questions for respiratory health, and eight questions that were applicable to both sleep and respiratory health (see Table 3).

## DISCUSSION

The RHSCN Research Prioritization process identified 17 priorities for respiratory and sleep health, integrating the perspectives of patients/caregivers and clinicians/researchers. These priorities included equitable access to health services, the economic impact of respiratory and sleep conditions, community-based management (such as referrals, self-management, when to seek medical care), and disease diagnosis, control and management (Table 3). This research is novel as it was co-owned and co-developed by patient/caregivers and clinicians/researchers, and is the first Canadian research

prioritization project that aimed to address both respiratory and sleep health. This prioritization will assist with decision making for the RHSCN related to allocation of resources and Network priorities.

Including individuals with personal experience as co-owners in research increases legitimacy and opportunity for innovation.[20] Additionally, researchers have moral and ethical responsibilities to ensure that the voice of patients and families are represented in research priorities.[21] A strength of this research is the high participation of patients and caregivers in the process, which is reflected in the nature of the research questions prioritized. As suggested by Breault and colleagues[14], the higher the level of participation from individuals with lived experiences (and their caregivers), the more closely a research prioritization list will reflect the true priorities of the community. The questions identified in this study had a strong focus on policy, equitable access to services, and the management of social, psychological, mental health issues related to respiratory/sleep health. Over two-thirds of survey respondents indicated they were patients, and almost half stated they were caregivers of someone with a respiratory or sleep problem (Note, individuals could select more than one role).

Respiratory-specific research prioritization has also increased in prominence. In the Netherlands, patient focus groups and questionnaires were used to identify research priorities for asthma and COPD.[22] Two significant priorities that arose were knowledge about the causes of the diseases and development of more effective medications, with little emphasis upon socio-political factors that impact health care. In contrast, our priorities for respiratory/sleep health research had a strong emphasis on social, economic and health care access issues. This variance may be the result of the autonomy of the Steering Committee, who made modifications to the proposed structure and inclusion criteria of the priorities by including out of scope policy and economic questions in the final review, or a reflection of shifting priorities over time, and the nature of the RHSCN being focused on health service delivery, rather than translational/clinical research.

Other targeted respiratory research prioritization activities identified some priorities with similar themes to this research. A formal JLA on asthma was previously conducted in the United Kingdom. [23] Our recent work is consistent with their JLA which identified the importance of managing adolescent and young adult care as an important priority, as well as identifying effective self-management interventions. Additional research has identified nursing-related respiratory research priorities through modified Delphi approaches.[24] This work determined patient understanding of asthma control and the clinical and cost-effectiveness of respiratory nurse interventions as the most prominent research priorities. [24] While these priorities were nursing-specific, cost/economic impact was highlighted as an important priority in this work as well. A separate modified Delphi was conducted in critical care, adult pulmonary, and sleep conditions across 45 countries to identify nursing research priorities to inform research and advise funding agencies. [25] This extensive project resulted in research priorities focused in four broad areas including: communication, education, risk reduction and psychological support. [25] Similar to this previous work, our project identified psychological factors that impact respiratory disease/sleep disorders and self-management supports (such as education) as important priorities.

In 2015, the American Thoracic Society released a statement on the importance of sleep health, and included research priorities. [26] This document highlighted the importance of conducting research linking sleep quality and health outcomes. [26] This priority, developed by international sleep medicine leaders is consistent with the research priority identified by our process (What strategies can be used to promote sleep as an important contributor to health?). A JLA initiative was recently completed in Saskatchewan, Canada to assess the research priorities for sleep apnea. [27] Alignment with our work shows common priorities related to equitable access, follow-up care/re-evaluation, and additional therapies (beyond CPAP) for OSA treatment. [27] Recent work in Quebec Canada surveyed 148 COPD patients to develop a health and healthcare research agenda for COPD. This work identified that relief of breathlessness was a research and health care priority for patients. (20). While management of

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breathlessness was not specifically identified within the current prioritization process, we did identify self-management and community supports/resources as a research priority (For individuals living with respiratory disease/sleep disorders, what are the most effective self-management interventions and/or community supports/resources?). Research prioritization activities have increased in recent years, and similarities with our work are demonstrated, however this research is important as it was developed by stakeholders from across the healthcare system with a focus on driving the local respiratory and sleep research agenda.

**LIMITATIONS**

There are limitations to consider related to this research. First, an online survey platform was convenient, cost-effective and timely; however, it may have prevented some individuals from participating in the research. Second, as participants self-selected their participation, our results may be influenced by motivation bias, selection bias, and participant sampling error. We did not undertake specific approaches to target Indigenous populations and caregivers, those with a primary language other than English, nor those with limited internet access. It is important to note that the study sample was predominantly female. These limitations of sampling methodology may limit the generalizability of study findings. In future research, we would advocate for requesting ethnicity/race information from participants and using this information to guide purposeful recruitment of participants. As well, we would suggest developing a targeted strategy for ensuring representation from those without internet access, and ensuring key populations are included.

Third, the time commitment for participating as a Steering Committee member was substantial. Members attended two full-day in-person meetings and approximately six hours of telephone meetings, plus numerous emails/material review over the course of 18 months. The participation level of the patient/caregiver steering committee demonstrated strong commitment, however future groups may consider additional strategies, such as role-sharing, to decrease volunteer fatigue. Also, these members

were identified specifically for participation in this work, which may have resulted in additional selection bias. Fourth, this research focused on the province of Alberta, and while the results may be broadly translatable, generalizability may be limited. However, prioritization activities driven by local stakeholders are important to providing research priorities of value to the local community. Finally, given the data for this study were collected over 2017-2019, there may be changes in respiratory research priorities following the COVID-19 pandemic.

## CONCLUSION

By involving individuals with personal experience with respiratory disease, caregivers and health care professionals, a research priority list comprising six questions for sleep health, three questions for respiratory health, and eight questions that were applicable to both sleep and respiratory health was developed. Broadly, this research was novel as the research priorities demonstrated an increased focus on issues such as equitable access, economic impact and community disease management (including resource allocation and self-management supports). This research exercise will lead to a more focused distribution of research funds and other resources locally, and will inform other groups as they look to support research that is more relevant to patients.



**Acknowledgements:** Our sincere appreciation to the Respiratory Health Section of the Medicine Strategic Clinical Network Core Committee for their ongoing support of this project. In addition, we thank all of the participants in the surveys and the research prioritization process. This work was supported by an in-kind contribution by the Respiratory Health Strategic Clinical Network™ of Alberta Health Services and the Alberta SPOR SUPPORT Unit Patient Engagement Platform.

**Funding statement:** This research received no specific grant from any funding agency in the public, commercial or not-for-profit sectors.

**Conflict of Interest Statement:** Ms. Hayward received honoraria for speaking at continuing education activities for health care professionals from Astra Zeneca in the last year, unrelated to this project. Dr. Sachin Pendharkar discloses he has received advisory fees from Jazz Pharmaceuticals, Paladin Labs and the International Centre for Professional Development in Health and Medicine; received research funding from Canadian Institute for Health Research, Alberta Health Services, The Lung Association, and MITACS; additionally, he has participated in a study by Jazz Pharmaceuticals (site co-investigator with no personal stipend or salary). Dr. Heather Sharpe & Dr. Michael Stickland received research funding from the Alberta Boehringer Ingelheim Collaboration and Dr. Sharpe is a member of the Board of Directors of the Canadian Network of Respiratory Care. The remaining authors have no other disclosures.

**Availability of data and materials-** the datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

**Ethics approval-** granted from the Health Research Ethics Board at the University of Alberta.

**Contributors’ Statement**

\*As this work was co-owned and developed in collaboration with our patient/caregiver advisors, all members of the Steering Committee were responsible for study design and interpretation of results. All authors have given permission for the submission of the manuscript, and have agreed to be listed as a co-author.

Heather Sharpe –led the Steering Committee (SC) through the study design process, data collection and analysis. Was responsible for drafting the manuscript, editing and is corresponding author.

Lisa Cerato-facilitated the final workshop to the creation of the final prioritization list. Provided feedback on the manuscript.

Darlene Derech-a patient/caregiver advisor that was a member of the SC. Involved in the study design, data collection, analysis and final research prioritization. Was responsible for providing feedback on the manuscript.

Lisa Guirguis-a health professional/academic representative on the SC. Involved in the study design, data collection, analysis and final research prioritization. Was responsible for providing feedback on the manuscript.

Kathy Hayward- a health professional/academic representative on the SC. Involved in the study design, data collection, analysis and final research prioritization. Was responsible for providing feedback on the manuscript.

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7 Joanna E. MacLean- a health professional/academic representative on the SC. Involved in the study  
8 design, data collection, analysis and final research prioritization. Was responsible for providing feedback  
9 on the manuscript.  
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11 Elizabeth Manafo-conducted the determination if submitted research questions were answered in the  
12 existing literature and responsible for the methodology and reporting. Provided feedback on the  
13 manuscript.  
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19 Sachin R. Pendharkar- a health professional/academic representative on the SC. Involved in the study  
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47 Mindy Tindall- a patient/caregiver advisor that was a member of the SC. Involved in the study design,  
48 data collection, analysis and final research prioritization. Was responsible for providing feedback on the  
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Harissios Vliagoftis- a health professional/academic representative on the SC. Involved in the study design, data collection, analysis and final research prioritization. Was responsible for providing feedback on the manuscript.

Michael K Stickland- the Scientific Director of the RHSCN. Involved in the study design, data collection, analysis and final research prioritization. Was responsible for providing feedback on the manuscript.

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Table 1. Survey participant demographic characteristics

Characteristic	No (%) of total respondents (n=448)
<b>Do you prefer to answer questions related to breathing problems, sleep problems or both? (n=409)</b>	
Breathing problems	81 (19.8)
Sleep problems	123 (30.1)
Both (breathing and sleep problems)	205 (50.1)
<b>Category-may choose more than 1 (n=446)</b>	
Person with breathing problems	132 (29.6)
Person with sleep problems	185 (41.5)
Family/caregiver of person with breathing problems	90 (20.2)
Family/caregiver of person with sleep problems	117 (26.2)
Health care professional-breathing problems	148 (33.2)
Health care professional-sleep problems	128 (28.7)
Other (dentist, scientist, patient with co-morbidities)	11 (2.5)
<b>Gender (n=446)</b>	
Female	347 (77.8)
Male	86 (19.3)
Non-Binary	5 (1.1)
Prefer not to respond/Did not respond	8 (1.8)
<b>Age</b>	
≤17 years	1 (0.2)
18-29 years	44 (9.9)
30-49 years	116 (26.0)
50-79 years	197 (44.2)
≥80 years	4 (0.9)
Did not respond	4 (0.9)

Table 2. Submitted Research Questions by Topic Reviewed and Finalized by Steering Committee

Total	Total questions	Answered in Literature	Remaining Research questions
Asthma	33	20	13
Combustibles*	4	1	3
COPD	3	2	1
Devices	13	4	9
Diagnostic	7	4	3
General Medicine	14	11	3
ILD	9	4	5
Nasal	4	3	1
OSA	25	19	6
Sleep	35	17	18
Other	25	4	21
Cost and policy**	0	0	0
Total	172	89	83

\*combustibles included cigarette smoking, vaping and cannabis, as well as secondary effects from these products.

\*\*cost and policy questions were deemed out of scope and represented in the out of scope data.

Table 3. List of Final Research Questions

<b>Questions for Respiratory and Sleep</b>	
1.	What is the relationship between social, psychological, and mental health issues and respiratory disease/sleep disorders for the individual/the family?
2.	What is the economic impact of respiratory disease/sleep disorders for community and inpatient/urgent care in Alberta?
3.	For individuals with suspected respiratory disease/sleep disorders, when is it appropriate for a primary care provider to refer to a specialist, compared to continuing care, to ensure the best treatment/health outcomes? What are the educational needs of primary care providers to facilitate this?
4.	What model of care is most effective at transitioning patients and their families from paediatric to adult respiratory/sleep care?
5.	For individuals living with respiratory disease/sleep disorders, what are the most effective self-management interventions and/or community supports/resources?
6.	What strategies will improve equitable access to respiratory/sleep care for Albertans?
7.	What are the patient & families' priorities related to the treatment of their breathing/sleep problems?
8.	For individuals with respiratory/sleep problems, which interventions, resources, programs in the community will result in fewer specialty care, emergency department or hospital visits?
<b>Questions for Respiratory</b>	
1.	How can we improve access to pulmonary rehabilitation for individuals with respiratory disease?
2.	For individuals with respiratory disease, how do environmental factors (humidity, air pollution etc.) impact disease control/management in Alberta?
3.	How does an individual with respiratory disease determine if different therapies will be worth the additional cost (money, risk of side effects, exacerbations)?
<b>Questions for Sleep</b>	
1.	For individuals with suspected sleep-related breathing disorders (e.g. obstructive sleep apnea), what is the recommended ongoing clinical management/follow-up care to improve and sustain health outcomes?
2.	Do treatments besides CPAP improve outcomes for individuals with sleep-related breathing disorders (e.g. obstructive sleep apnea and/or hypoventilation)?
3.	What is the current quality of provision and interpretation of investigations for sleep-related breathing disorders in Alberta?
4.	What strategies can be used to promote sleep as an important contributor to health?
5.	For individuals experiencing poor quality sleep, at what point should they seek medical advice to improve their health outcomes?
6.	What are the policy factors that inform insurable coverage for testing and treatment of sleep-related breathing disorders in Alberta?

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Appendix A. Verification Process to Identify Uncertainties

Where, no up-to-date, reliable systematic reviews or guidelines of research evidence addressing the uncertainty exist/have not been adequately addressed. If a study protocol exists, this will be noted. These uncertainties will move forward for interim prioritization.

Process	Description	Output
a. Check evidence-base for relevant, up to date systematic reviews, guidelines and study protocols (i.e., studies in process)	<ul style="list-style-type: none"><li>i. Cochrane Database of Systematic Reviews (EBM Reviews)</li><li>ii. American College of Physicians (ACP) Journal Club via Medline (Ovid)</li><li>iii. Database of Abstracts of Reviews of Effects (DARE) via Medline (Ovid)</li><li>iv. Medline databases (Ovid)</li><li>v. <a href="#">Canadian Respiratory Guidelines</a> (Canadian Thoracic Society)</li><li>vi. <a href="#">Global Initiative for Asthma</a> (GINA)</li><li>vii. <a href="#">AAFP</a> (USA)</li><li>viii. <a href="#">NICE guidelines</a> (UK)</li><li>ix. Other (used if unique population, intervention or outcome would be better addressed in a different database)</li></ul>	If evidence is available, cite it with primary author conclusion.
	Using specific search terms as they relate to the submission topic area, subtopic area and outcome (as identified). Specific search outputs will be documented as they relate to each topic and subtopic area.	
	To check questions about sleep and breathing risk factors, which are mainly addressed by reviews including prospective cohort studies, other systematic reviews. MEDLINE, EMBASE, CINAHL and PsycINFO will be searched with the same keywords using a systematic review filter.	
b. Determine if evidence is reliable and relevant using	1] Systematic review	
	1.1 Has it been published in the last 3 years?	[YES > does not move forward; NO > moves forward]

Process	Description	Output
the following criteria:	<p><b>AND</b></p> <p>1.2 For evidence not derived from Cochrane et al., ask the following: Does it have published clear methods?</p> <ul style="list-style-type: none"> <li>• Where inclusion/exclusion criteria reported?</li> <li>• Was search adequate?</li> <li>• Where the included studies synthesized?</li> <li>• Was the quality of included studies assessed?</li> <li>• Are sufficient details about the individual included studies presented?</li> <li>• Was any potential bias addressed/managed?</li> </ul>	[YES > does not move forward; NO > moves forward]
	<p><b>AND</b></p> <p>1.3 Are reported findings clinically significant?</p>	[YES > does not move forward; NO > moves forward]
	<b>2] Guidelines</b>	
	2.1 Are guidelines evidence-based, that are used as a framework for clinical decision making and supporting best practices from authoritative Canadian, American or UK bodies?	[YES > does not move forward; NO > moves forward]
	<b>3] Protocol</b>	
	3.1 If no systematic review or guidelines have been located, conduct Protocol search through EBM Cochrane	[YES > note its existence; NO > moves forward]
		<p>If <b>all of the criteria</b> have been answered with a <b>Yes = "Known unknown"</b> [tier 2]</p> <p>If <b>one or more</b> of the criteria has been answered with a <b>No = Uncertainty</b></p>

Process	Description	Output
c. Verify selection output	Clinical significance must be determined by Steering Committee based on reported findings and recorded author conclusions.	Long-list of uncertainties are identified for interim prioritization (and 'known unknowns' are recorded with evidence cited)

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### Reporting guideline for health research priority setting with stakeholders (REPRISE)

No	Item	Descriptor and/or examples
A	Context and scope	
1	Define geographical scope	Provincial scope
2	Define health area, field, focus	Respiratory and sleep health
3	Define the intended beneficiaries	Individuals living with respiratory and sleep disorders in the province of Alberta and their families.
4	Define the target audience of the priorities	Respiratory Health Section of the Medicine Strategic Clinical Network-to use the prioritization to fund research
5	Identify the research area	Health service research and clinical research
6	Identify the type of research questions	Was not pre-defined
7	Define the time frame	To provide research prioritization for five years
B	Governance and team	
8	Describe the selection and structure of the leadership and management team	The Scientific Office of the Network was primarily responsible for the development of the Steering Committee. The Steering Committee was created to ensure a strong voice for those with lived experience of respiratory and/or sleep disorders. The Core Committee of the Network provided content expertise.
9	Describe the characteristics of the team	Project oversight was performed by a Steering Committee comprising four individuals with lived experience, two caregivers, four respiratory physicians,

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No	Item	Descriptor and/or examples
		two respiratory therapists, two pharmacists, one nurse and the Scientific Director of the RHSCN, within the group there was representation from academic and community based clinicians. We did not request Steering Committee members to disclose sex/gender or race/ethnicity, however the group was diverse in perceived age, sex, gender, ethnicity and academic/non-academic background.
10	Describe any training or experience relevant to conducting priority setting	The Patient Engagement Platform of the Alberta Strategy for Patient-Oriented Research's Support for People and Patient-Oriented Research and Trials Unit provided guidance, a consultant with priority setting experience was hired to assist with identifying unanswered questions and a facilitator was brought to the team for the final priority setting workshop.
C	Framework for priority setting	
11	State the framework used (if any)	A modified James Lind Alliance approach was used based upon the Alberta Depression Research Priority Setting Project
D	Stakeholders or participants	
12	Define the inclusion criteria for stakeholders involved in priority-setting	Patients, caregivers, health professionals, researchers, policy makers, and other stakeholders that self-identified as having an interest in respiratory/sleep health
13	State the strategy or method for identifying and engaging stakeholders	Posters and cards with QR codes and website links were disseminated through stakeholders, such as respiratory and sleep clinics. Steering Committee members were asked to share the flyer within their community (such as support groups, online forums etc.) and through social media platforms. The

No	Item	Descriptor and/or examples
		RHSCN shared the survey links through newsletters and email communications to members.
14	Indicate the number of participants and/or organizations involved	A total of 461 unique individuals accessed the first survey. Fifteen participants' responses were removed as they did not provide clear consent, leaving a total of 446 survey responses.
15	Describe the characteristics of stakeholders	Three core groups: 317 participants stated they were an individual with lived experience with respiratory disease/patient (71.1%), 207 stated they were a caregiver of an individual with respiratory disease (46.4%) and 276 stated they were a health care professional that provided care for individuals with respiratory disease (61.9%). Of note, many respondents selected more than one category with which they identified.
16	State if reimbursement for participation was provided	Individuals that participated on the Steering Committee were provided reimbursement for any expenses associated with attending the in-person meetings including travel (hotel, mileage, parking, meals etc.). Participants were not paid an honoraria or other compensation. Participants in the survey were invited to enroll in a draw for a small gift card to a book store for participating.
E	Identification and collection of research priorities	
17	Describe methods for collecting initial priorities	The four phases to the project included (1) identifying research questions by stakeholders, (2) determining research questions that had been answered in existing literature (guidelines, systematic reviews etc.), (3) prioritization of unanswered questions, and (4) finalization through an in-person priority setting workshop. The Steering Committee designed the initial survey asking



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No	Item	Descriptor and/or examples
		two open-ended core questions, (1) What questions do you have about breathing problems? And (2) What questions do you have about sleep problems?
18	Describe methods for collating and categorizing priorities	Priorities were originally organized into the broad categories of respiratory health and sleep health. Subsequently they were categorized into sub-groups to facilitate data management. For example, respiratory health priorities were organized into: asthma, COPD, medications, devices, obstructive sleep apnea, cost & policy, diagnostic, nasal, sleep, interstitial lung disease, and other.
19	Describe methods and reasons for modifying (removing, adding, reframing) priorities	All proposed priorities were reframed as necessary to create a PICO (patient, intervention, control & outcome) question were possible. Priorities that overlapped were combined.
20	Describe methods for refining or translating priorities into research topics or questions	Two members of the team were responsible for creating research questions from the submissions. The resulting list was reviewed by the Steering Committee.
21	Describe methods for checking whether research questions or topics have been answered	For each question topic, key databases were searched for relevant, up-to-date systematic reviews, clinical practice guidelines and/or study protocols (i.e., studies in process). Questions that had not been adequately answered in the literature (the topic was not addressed by the above sources), were to be included in the list of unanswered questions. Questions that were answered in the literature were identified as 'known unknowns', and along with possibly out of scope questions (policy questions or those that would not be answered by the literature sources) were included by the two reviewers in a separate list for review by list to the Steering Committee. Questions for final inclusion

No	Item	Descriptor and/or examples
		based on clinical and research significance were determined by the Steering committee.
22	Describe number of research questions or topics	A total of 595 research questions were submitted (see Figure 1). A team of two people identified answers for 343 (74%) questions within the literature. A total of 178 research questions were identified as unanswered questions that moved forward. One hundred forty stakeholders participated in the second survey to rank the remaining questions. A final list of 17 research topics was identified as priority areas for respiratory and sleep research in Alberta through this prioritization.
F	Prioritization of research topics/questions	
23	Describe methods and criteria for prioritizing research topics or questions	Two online surveys were completed. The first, to identify possible priorities, the second to prioritize the identified priorities from the first survey. The Network Core Committee reviewed the results and provided their input to the Steering Committee. An in person priority setting workshop was completed to identify the final priorities.
24	State the method or threshold for excluding research topics/questions	A consensus approach was used. The Steering Committee reached consensus on what questions would be included in the final list.
G	Output	
25	State the approach to formulating the research priorities	PICO (population, intervention, comparator, outcome) questions were formulated, however the Steering Committee did revise the priorities in the final priority setting workshop.
H	Evaluation and feedback	

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No	Item	Descriptor and/or examples
26	Describe how the process of prioritization was evaluated	The prioritization process has not yet been formally evaluated, however the impact will be assessed through the Scientific Office of the Network.
27	Describe how priorities were fed back to stakeholders and/or to the public; and how feedback (if received) was addressed and integrated	The results of the priority setting were shared with the RHSCN, including the Core Committee and the working groups.
I	Implementation	
28	Outline the strategy or action plans for implementing priorities	The priorities have been used to guide the funding of research grants offered by the Network, including seed grant funding and studentships.
29	Describe plans, strategies, or suggestions to evaluate impact	The Network evaluates the impact of the scientific office annually, as well as is involved in the evaluation and priority setting for the Network every 3-5 years.
J	Funding and conflict of interest	
30	State sources of funding	This work was supported by an in-kind contribution by the Respiratory Health Strategic Clinical Network™ of Alberta Health Services and the Alberta SPOR SUPPORT Unit Patient Engagement Platform.
31	Declare any conflicts or competing interests	The authors have no other disclosures.

### Checklist for Reporting Results of Internet E-Surveys (CHERRIES)

<b>Checklist Item</b>	<b>Explanation</b>	<b>Page Number</b>
Describe survey design	Describe target population, sample frame. Is the sample a convenience sample? (In “open” surveys this is most likely.)	Page 6-7
IRB approval	Mention whether the study has been approved by an IRB.	Page 7
Informed consent	Describe the informed consent process. Where were the participants told the length of time of the survey, which data were stored and where and for how long, who the investigator was, and the purpose of the study?	Page 7
Data protection	If any personal information was collected or stored, describe what mechanisms were used to protect unauthorized access.	n/a
Development and testing	State how the survey was developed, including whether the usability and technical functionality of the electronic questionnaire had been tested before fielding the questionnaire.	Page 7
Open survey versus closed survey	An “open survey” is a survey open for each visitor of a site, while a closed survey is only open to a sample which the investigator knows (password-protected survey).	Page 7
Contact mode	Indicate whether or not the initial contact with the potential participants was made on the Internet. (Investigators may also send out questionnaires by mail and allow for Web-based data entry.)	Page 7
Advertising the survey	How/where was the survey announced or advertised? Some examples are offline media (newspapers), or online (mailing lists – If yes, which ones?) or banner ads (Where were these banner ads posted and what did they look like?). It is important to know the wording of the announcement as it will heavily influence who chooses to participate. Ideally the survey announcement should be published as an appendix.	Page 6
Web/E-mail	State the type of e-survey (eg, one posted on a Web site, or one sent out through e-mail). If it is an e-mail survey, were the responses entered manually into a database, or was there an automatic method for capturing responses?	Page 6
Context	Describe the Web site (for mailing list/newsgroup) in which the survey was posted. What is the Web site about, who is visiting it, what are visitors normally looking for? Discuss to what degree the content of the Web site could pre-select the sample or influence the results. For example, a survey about vaccination on a anti-immunization Web site will have different results from a Web survey conducted on a government Web site	Page 6
Mandatory/voluntary	Was it a mandatory survey to be filled in by every visitor who wanted to enter the Web site, or was it a voluntary survey?	Page 6
Incentives	Were any incentives offered (eg, monetary, prizes, or non-monetary incentives such as an offer to provide the survey results)?	Page 6

Time/Date	In what timeframe were the data collected?	Page 6
Randomization of items or questionnaires	To prevent biases items can be randomized or alternated.	n/a
Adaptive questioning	Use adaptive questioning (certain items, or only conditionally displayed based on responses to other items) to reduce number and complexity of the questions.	n/a
Number of Items	What was the number of questionnaire items per page? The number of items is an important factor for the completion rate.	Page 7
Number of screens (pages)	Over how many pages was the questionnaire distributed? The number of items is an important factor for the completion rate.	Page 7
Completeness check	It is technically possible to do consistency or completeness checks before the questionnaire is submitted. Was this done, and if “yes”, how (usually JavaScript)? An alternative is to check for completeness after the questionnaire has been submitted (and highlight mandatory items). If this has been done, it should be reported. All items should provide a non-response option such as “not applicable” or “rather not say”, and selection of one response option should be enforced.	n/a
Review step	State whether respondents were able to review and change their answers (eg, through a Back button or a Review step which displays a summary of the responses and asks the respondents if they are correct).	n/a
Unique site visitor	If you provide view rates or participation rates, you need to define how you determined a unique visitor. There are different techniques available, based on IP addresses or cookies or both.	n/a
View rate (Ratio of unique survey visitors/unique site visitors)	Requires counting unique visitors to the first page of the survey, divided by the number of unique site visitors (not page views!). It is not unusual to have view rates of less than 0.1 % if the survey is voluntary.	n/a
Participation rate (Ratio of unique visitors who agreed to participate/unique first survey page visitors)	Count the unique number of people who filled in the first survey page (or agreed to participate, for example by checking a checkbox), divided by visitors who visit the first page of the survey (or the informed consents page, if present). This can also be called “recruitment” rate.	n/a
Completion rate (Ratio of users who finished the survey/users who	The number of people submitting the last questionnaire page, divided by the number of people who agreed to participate (or submitted the first survey page). This is only relevant if there is a separate “informed consent” page or if the survey goes over several pages. This is a measure for attrition. Note that	n/a

agreed to participate)	“completion” can involve leaving questionnaire items blank. This is not a measure for how completely questionnaires were filled in. (If you need a measure for this, use the word “completeness rate”.)	
Cookies used	Indicate whether cookies were used to assign a unique user identifier to each client computer. If so, mention the page on which the cookie was set and read, and how long the cookie was valid. Were duplicate entries avoided by preventing users access to the survey twice; or were duplicate database entries having the same user ID eliminated before analysis? In the latter case, which entries were kept for analysis (eg, the first entry or the most recent)?	n/a
IP check	Indicate whether the IP address of the client computer was used to identify potential duplicate entries from the same user. If so, mention the period of time for which no two entries from the same IP address were allowed (eg, 24 hours). Were duplicate entries avoided by preventing users with the same IP address access to the survey twice; or were duplicate database entries having the same IP address within a given period of time eliminated before analysis? If the latter, which entries were kept for analysis (eg, the first entry or the most recent)?	n/a
Log file analysis	Indicate whether other techniques to analyze the log file for identification of multiple entries were used. If so, please describe.	n/a
Registration	In “closed” (non-open) surveys, users need to login first and it is easier to prevent duplicate entries from the same user. Describe how this was done. For example, was the survey never displayed a second time once the user had filled it in, or was the username stored together with the survey results and later eliminated? If the latter, which entries were kept for analysis (eg, the first entry or the most recent)?	n/a
Handling of incomplete questionnaires	Were only completed questionnaires analyzed? Were questionnaires which terminated early (where, for example, users did not go through all questionnaire pages) also analyzed?	Page 8
Questionnaires submitted with an atypical timestamp	Some investigators may measure the time people needed to fill in a questionnaire and exclude questionnaires that were submitted too soon. Specify the timeframe that was used as a cut-off point, and describe how this point was determined.	n/a
Statistical correction	Indicate whether any methods such as weighting of items or propensity scores have been used to adjust for the non-representative sample; if so, please describe the methods.	n/a

This checklist has been modified from Eysenbach G. Improving the quality of Web surveys: the Checklist for Reporting Results of Internet E-Surveys (CHERRIES). J Med Internet Res. 2004 Sep 29;6(3):e34 [erratum in J Med Internet Res. 2012; 14(1): e8.]. Article available at <https://www.jmir.org/2004/3/e34/>; erratum available <https://www.jmir.org/2012/1/e8/>. Copyright ©Gunther Eysenbach. Originally published in the [Journal of Medical Internet Research](https://www.jmir.org/2004/3/e34/), 29.9.2004 and 04.01.2012.

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